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# THE JOURNAL OF PEDIATRICS

A MONTHLY JOURNAL DEVOTED TO THE PROBLEMS AND DISEASES OF INFANCY AND CHILDHOOD

BORDEN S. VEEDER
St. Louis
*Editor*

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Clinical evidence indicates that the immunity produced by combined immunization against diphtheria and tetanus is at least as effective as that obtained with single immunization preparations. Combined vaccines have the added advantage of reducing the total number of injections required. Physicians now may virtually strike out two scourges of infancy and early childhood with a single preparation. Diphtheria Toxoid-Tetanus Toxoid Combined, Alum Precipitated, is available in packages containing two 1-cc. vials (1 immunization), and in 10-cc. vials (5 immunizations).

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# The Journal of Pediatrics

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## Original Communications

### THE ULCERATED URETHRAL MEATUS IN MALE CHILDREN


PAUL FREUD, M.D.
NEW YORK, N. Y.


SUPERFICIAL ulceration of the urethral meatus is a definite entity occurring in male infants and young boys, which, in spite of its frequent incidence, is little known. It occurs chiefly in circumcised children but may be found in the noncircumcised as well if the prepuce leaves the tip of the glans free to irritation.

This peculiar lesion consists of a superficial ulcer affecting the periphery of the meatus and cracking its labia to a width of up to 3 mm. and its depth to about the same extent. The ulcer always remains superficial and localized and never involves the deeper parts of the urethra. Urethral discharge never occurs and there is no fever. The inguinal lymph nodes are sometimes swollen and tender. Crusts form rapidly on the surface of the ulcer and may partially or completely obstruct the meatus. This leads to difficulty in voiding and therefore is followed by urinary retention.

In many of these cases the children are unable to urinate before the obstructing crust has been removed either by pressure of overflowing urine or mechanically by parent, nurse, or physician. Voiding may be deferred from ten to twelve hours.

If the orifice is obstructed by scabs, the bladder will be only incompletely emptied by a divided stream or dribbling. When the crusts come off, the first drop of urine that touches the surface of the ulcer irritates and causes intense pain. The children, therefore, refuse to urinate until an overflowing bladder forces voiding. Following the passage of a few drops of urine, pain diminishes and enables the children to urinate. As soon as a scab comes off, bleeding occurs from the ulcer and bright bloody spots on the diapers or pants are frequently the first sign that alarms the parents.

Secretion on the surface of the ulcer soon starts again after the scabs have come off and new crusts are formed, causing the same difficulties and bleeding when they are removed. This vicious cycle may repeat itself many times. Eventually the ulcer heals spontaneously with resulting small constricting scars. After an interval of some time the disorder recurs and, if not treated,


From the Department of Pediatrics, New York Medical College, Flower and Fifth Avenue Hospitals, and the Metropolitan Hospital of the City of New York.

heals spontaneously, but again with the formation of scars and an increase of the superficial stricture. Any stricture of the urethra located in the orifice or at higher parts will eventually lead to retrograde pressure and distention with all its after-consequences of infection and pressure-damage.

The disorder was first seen by J. von Bokay, Sr.[1] in 1878 in twenty-three boys that were all Jewish and all circumcised. He called the lesion "urethritis orificii externi." He believed that the condition was due to poor hygiene but expressed the opinion that it may also be due to certain irritating properties of the urine. F. R. Sturgis[2] gave the first description of the entity in American literature in 1890. H. Goldenberg,[3] in 1910, presented one case describing the lesion in detail, but without consideration of etiology and treatment. T. S. Southworth[4] believes trauma and ammoniacal urine to be the etiologic factors. J. Zahorsky,[5] in a paper on diaper rash, mentions the disorder and believes the skin condition to be the essential etiologic factor. J. V. Cooke [6, 18] describes the condition in an article on ammonia dermatitis, and in his chapter on the same subject in Brennemann's *Practice of Pediatrics*. A very thorough paper on the condition was published by Brennemann[7] in 1921. He believed at first that the lesion was due to the most accessible and vulnerable part of the glans rubbing against clothing, but he could not explain the extension of the ulcer into the urethra by trauma alone. Since he observed similar lesions appearing simultaneously about the diaper region in many of these cases, he agreed with Zahorsky that "ammonia dermatitis" (diaper rash) should be considered the etiologic factor. In 1922, J. von Bokay, Jr.[8] reported a case in a 13-year-old noncircumcised boy, and thought that the lesion in this boy was brought about by masturbation. He also gave a list of 166 cases that were observed during a period of thirty-seven years at his hospital. Of these, thirty-eight were up to 1 year of age, ninety-four from 1 to 3 years, twenty-five from 3 to 7 years, and nine from 7 to 14 years. A. L. Wolbarst,[9] in his article on stenosed urinary meatus and its treatment, saw the disorder occur when infections and inflammatory swelling further diminished an already tight meatus. He recommends meatotomy and stresses that even this operation has to be followed by dilatations for several weeks to prevent the possibility of recontraction. In 1932, M. F. Campbell[10] published a case of congenital atresia of the meatus in a 7-year-old boy with pyuria and hematuria, and presented the x-ray pictures showing marked dilatation of the urethra and a hypertrophic bladder. The same author,[11] in his book on pediatric urology in 1937, and in his chapter on the male reproductive system in Brennemann's *Practice of Pediatrics* states that he never observed a case where diaper rash alone was responsible for the condition but that only boys with a congenitally tight meatus developed the lesion. An article on coarctation of the meatus was published by B. S. Abeshouse and D. E. Bogorad[12] in 1938. These authors presented thirteen cases of patients ranging from 7 months to 7 years of age, twelve of them circumcised and mostly Jewish; one with a long pendulous foreskin leaving the tip of the glans free to irritation. They consider the ammoniacal urine, discussing metabolic disturbances as its reason and gastrointestinal upsets as possible etiologic agents and are not in accord with Campbell's contention that a congenitally small meatus is the chief predisposing factor.

W. Hofmann,[13] in 1940, described a case of ulcerated meatus in a circumcised infant. In this case, however, the stenosis was not congenital but due to postoperative adhesions following the circumcision. In his paper on stenosis of the external urethral meatus, M. F. Campbell[14] states, in 1943, that 98 per cent of the stenosed cases are congenital. As a complication of this stenosis, ulceration occurs when and if inflammatory swelling narrows the stenosed orifice still more. After a description of the clinical criteria, he stresses the fact that, in spite of the generally accepted opinion, this disorder should not be considered as benign since urinary back pressure damages the proximal urinary tract and particularly the kidney. In sixty-three out of his 152 patients that ranged in age from 7 weeks to 16 years, urographic findings were abnormal. He recommends liberal meatotomy followed by "periodic progressive dilatation" of the stenosed meatus and treats coexisting infections with urinary antiseptics.

It is astonishing how few articles on this frequent and important disorder have been written and how little the condition is known. Apart from the discussions in Brennemann's *Pediatrics*,[15] the subject is hardly mentioned in pediatric textbooks. J. P. C. Griffith and A. G. Mitchell[17] speak about an inflammation of the external urethral orifice probably due to ammoniacal urine. C. Noeggerat and A. Eckstein,[18] in Pfaundler and Schlossmann's Handbuch Der Kinderheilkunde, give a short description of the disorder.

## CASE REPORTS

Twenty-two of the twenty-five cases listed in the table were observed in the pediatric clinics of the Flower and Fifth Avenue Hospitals and the Metropolitan Hospital in New York, at regular intervals for a period of at least one and a half years. The infants were of Jewish, Irish, Italian, Puerto Rican, and Negro extraction. Three of them were breast fed (Cases 3, 10, and 18). Since relapses of the condition may occur after 5 to 8 months of symptom-free intervals, it is only possible to regard a patient as cured after a long period of observation. Therefore, sixteen more patients, ranging in age from 5½ months to 7 years, with ulcerated meatus, seen at these clinics, are not listed in this table because the mothers failed to bring them back for regular check-ups.

All of the tabled and thirteen of the nonlisted cases showed a shortening of the anterior-posterior diameter of the urethral orifice to 3 mm. or less. In severe cases, only a pin-point opening could be seen. Several patients (Cases 15, 16, and 19) were seen first only with a congenital shortening of the anterior-posterior diameter and no other pathognomonic signs, and later on came back to the clinic with the typical picture of an ulcerated meatus. There was first an erythema involving the tip of the glans, then small vesicles appeared at the margin of the external meatus. Their rupture left small ulcerations that eventually became confluent. They were situated at the mucocutaneous border surrounding the meatus for a diameter of 2 to 4 mm. These ulcerations were covered by a gray scab that was so firmly adherent that its removal caused pain and bleeding. Due to these scabs, the adjacent labia of the meatus became firmly attached and obstruction was produced. Only after a certain time of obstruction the mucous

membrane of the urethra became inflamed and superficial ulcerations occurred that extended to a depth of up to 3 mm.

The healing of the ulcers was rather rapid, with the formation of scars. These scars, appearing as small rhagades at the lateral urethral labia of the meatus, were readily seen later on when there was no ulceration or inflammation.

In the majority of the tabled cases and in eleven of the cases not listed, the anterior posterior diameter was not much shorter than 3 mm. [Cases 1, 2, 3, 5, 6 (Fig. 5), 7 (Fig. 7), 8 (Fig. 6), 9, 10, 12, 13, (Fig. 3), 14 (Fig. 1), 20 (Fig. 2), 21 and 22]. In Cases 4, 15, 16, and 18, the anterior-posterior diameter of the orifice was so short that the babies had to be referred to the Urological Clinic, where successful dilatation by bougie was done. In Cases 11 (Fig. 4), 17, 19, 23, 24, and 25 (the last three observed in 1946), there was only a pin-point opening. These cases had to undergo meatotomy.

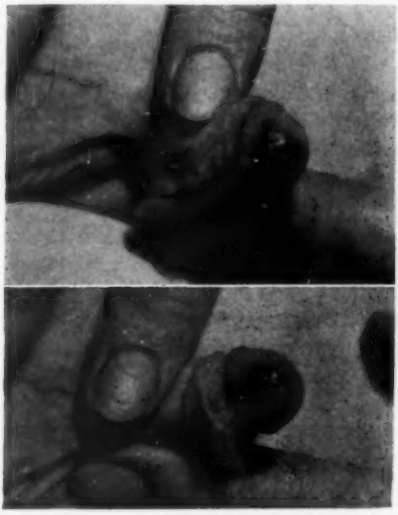


Fig. 1.—*A*, Case 14, L. H., showing the ulcerated meatus and dermatitis localized to the glans, leaving thighs and scrotum entirely free. *B*, Case 14, showing ulceration coarctation.

The lesion occurred usually in circumcised boys. In cases 6, 9, 10 and 24 the glans was left free to irritation by a wide pendulous foreskin. Case 9 was a boy with hypospadias and ulcerated meatus. This combination has frequently been seen by M. F. Campbell.[11]

In the majority of the cases, ammonia dermatitis was seen as an etiologic skin factor (Cases 1, 4, 5, 7, 8, 10, 12, 13, 15, 16, 19, 21, 22, 23, and 24). Twelve of the fifteen cases not listed showed the same condition. Faulty diet as a probable cause of this dermatitis (too much orange juice, too much cream, butter, or milk) was reported by the mothers of Cases 1, 8, 15, 21, 23, and 24. Ammonia smell and high pH factors of the urine were found in a number of these cases of diaper rash.

Six patients presented a peculiar dermatitis, which affected only the surroundings of the meatus and parts of the glans. This dermatitis left the skin of the scrotum, thighs, and abdomen entirely free. It consisted of reddened areas that were slightly raised and showed a roughened surface. In none of these cases could ammonia smell from the urine ever be detected, nor was the urine pH increased. Whether a fungus or allergy was responsible for this peculiar dermatitis or whether it was due to other unknown factors could not be determined. In Cases 2, 6, and 11, a typical herpes simplex of the penis was found to be the skin factor. Cultures from the orifice frequently showed *Staphylococcus albus* or *aureus*, but this should not be considered in any way conclusive, since it is almost impossible to avoid contamination of the cultures in this area. The patients ranged in age from 3 months to 7 years. Ulceration, crust formation, difficulty and pain in voiding, and bleeding were the constant signs and symptoms.

Fig. 2.

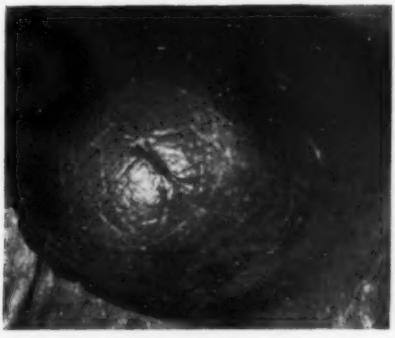


Fig. 3.

Fig. 2.—Case 20, T. G., showing ulcerated meatus and dermatitis localized to glans only.
Fig. 3.—Case 13, V. G., showing congenitally small meatus. No fissures on lateral labia. Beginning reddening and swelling. About two and one-half times normal size.

## TREATMENT

The acute condition with difficulty and pain in voiding is best treated according to Brennemann's proposal, by a drop of 2 per cent novocain solution on the surface of the ulcers, which immediately relieves pain and enables the children to urinate. The mothers are told to place the children in a warm bath which stimulates voiding and relieves pain by diluting the escaping urine. The formation of crusts after the removal of a scab can best be prevented by the frequent application of petrolatum or a 5 per cent boric acid lanolin ointment. This is, however, not sufficient in cases where reddening and swelling indicate an acute infection. Here, 5 per cent sulfathiazole ointment in lanolin base or a 5 per cent penicillin ointment have done well.

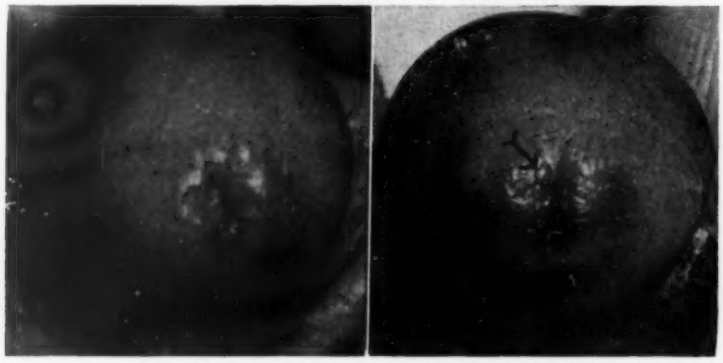
The cure of the contributing skin factor alone is insufficient as long as the anterior posterior diameter is not lengthened to its normal extent, which in children is 3 mm. or somewhat more. Wherever this has not been attained the condition will recur. In these recurring cases, especially if there are any signs of infection of the genitourinary tract, urography should be done.

TABLE 1

| CASE | NAME | AGE | CIRCUMCISED | LENGTH OF ANTERO-POSTERIOR DIAMETER | | | CRUST FORMATION | DIFFICULTY IN VOIDING | BLEEDING | FAULTY DIET | AMMONIA DERMATITIS | DERMATITIS AFFECTING GLANS OR MEATUS ONLY | HERPES SIMPLEX | TREATMENT | | | REMARKS |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | 1-3 MM. | ABOUT 1 MM. | PIN POINT | | | | | | | | MICROCUETIP DILATATION | SOUND DILATATION | MEATOTOMY | |
| | | | | | | | | | 1941 | | | | | | | | |
| 1 | G. J. | 2 yr. | + | + | | | + | + | + | + | + | | | + | | | 8 oz. orange juice per day substituted by 50 mg. ascorbic acid; two relapses. |
| 2 | M. L. | 13 mo. | + | + | | | + | + | | | | | + | + | | | Three relapses. |
| 3 | S. J. | 3 mo. | + | + | | | + | + | + | | | + | | + | | | One relapse. |
| 4 | R. L. | 7 mo. | + | | + | | + | + | + | | + | | | | + | + | Repeated dilatations with bougie, eventually meatotomy. |
| 5 | E. R. | 5 mo. | + | + | | | + | + | + | | + | | | + | | | Two relapses. |
| 6 | S. C. | 5 yr. | No | + | | | + | + | + | | | | + | + | | | Three relapses. |
| 7 | J. L. | 2½ yr. | + | + | | | + | | + | | + | | | + | | | Four recurrences. |
| | | | | | | | | | 1942 | | | | | | | | |
| 8 | W. A. | 8 mo. | + | + | | | + | + | + | + | + | | | + | | | 10 oz. orange juice, plenty of butter and cream daily. |
| 9 | J. W. | 2½ yr. | No | + | | | + | + | | | | + | | + | | | Hypospadia, frequent ulceration. |
| 10 | D. B. | 3 mo. | No | + | | | + | + | + | | + | | | + | | | Pendular foreskin; four relapses. |
| 11 | A. C. | 8 mo. | + | | | + | + | + | + | | | | + | | | + | Several relapses of herpes but no relapses of ulcers after meatotomy. |

| | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 12 | G. A. | 6 mo. | + | + | | | + | + | + | | + | | | + | | Ulceration occurred twice. |
| 13 | V. G. | 20 mo. | + | + | | | + | + | + | | + | | | + | | No relapse. |
| 14 | L. H. | 20 mo. | + | + | | | + | + | + | | | + | | + | | No relapse. |
| | | | | | | | | 1943 | | | | | | | | |
| 15 | R. W. | 18 mo. | + | | + | | + | + | + | + | + | | | | + | Plenty of cream and butter daily; one relapse. |
| 16 | J. A. | 18 mo. | + | | + | | + | + | + | | + | | | | + | No relapse. |
| 17 | L. S. | 2 yr. | + | | | + | + | + | + | | | + | | | + | + | Meatotomy after several unsuccessful sound dilatations. |
| 18 | R. J. | 5 mo. | + | | + | | + | | + | | | + | | | + | | Cured after several sound dilatations. |
| 19 | M. S. | 2 yr. | + | | | + | + | + | + | | + | | | | + | + | Several dilatations unsuccessful, cured after meatotomy despite recurring diaper rash. |
| 20 | T. G. | 11 mo. | + | + | | | + | | + | | | + | | + | | | One brother was observed with the same condition. |
| | | | | | | | | 1944 | | | | | | | | |
| 21 | J. S. | 15 mo. | + | + | | | + | + | + | + | + | | | + | | | 2 quarts of milk daily, diet advice; two relapses. |
| 22 | J. K. | 2½ yr. | + | + | | | + | + | + | | + | | | + | | | No relapse. |
| | | | | | | | | 1946 | | | | | | | | |
| 23 | A. A. | 5 mo. | + | | | + | + | + | + | + | + | | | | | + | 8 oz. orange juice per day, cured after meatotomy. |
| 24 | F. P. | 5 yr. | No | | | + | + | + | | + | + | | | | | + | Plenty of cream daily. Meatotomy. Diaper rash recurring, no more ulceration. |
| 25 | R. McM. | 22 mo. | + | | | + | + | + | + | | | + | | | | + | First attack. |

The majority of the presented cases showed only a moderate shortening of this diameter and ammonia dermatitis. A method which is effective in these cases is to insert, once daily, "microcuetips" (toothpicks prepared in a manner of ear swabs with cotton projecting one-half inch beyond the tip of the toothpick). The mothers were told to put the cotton end into the prescribed ointment and to insert it into the meatus to the depth of about 5 mm. once daily for a



*A.* *B.*

Fig. 4.—*A*, Case 11, A. C., first attack. Multiple fissures around orifice. Small ulcer near dorsal end of opening. *B*, second attack. Multiple small fissures on lateral labia with constriction of orifice. Blister on right labium (arrow). About two and one-half times normal size.

Fig. 5.—Case 6, S. C. Irregularly constricted orifice due to many small labial scars. Ulcer on right labium. Intensive swelling of left labium. About two and one-half times normal size.

period of three months. In these cases, this treatment was sufficient to extend the anterior-posterior diameter to its normal physiological length of about 3 mm. or more. In any case of ammonia dermatitis it is just as important to cure this condition and to prevent its recurrence. The diapers should be treated with a nonvolatile antiseptic. This will prevent the occurrence of ammonia dermatitis. In greater degrees of shortening, bougie dilatation was done in the

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Fig. 6.—Case 8, W. A. Inflammatory reddening and swelling with ulceration of the meatus.

urological clinic and in the relatively rare cases where there was only a pinpoint opening, meatotomy had to be performed.

In the majority of the cases where the skin factor was ammonia dermatitis, the following steps were suggested. The diapers were washed in saturated boric acid solution instead of soap and were not rinsed to obtain the best possible change of the pH from alkaline to acid. Wherever the mothers could afford it, modern neutral soaps were recommended for bathing the children. Otherwise, only boric acid solution was used for this purpose. One to two per cent tar in zinc oxide paste was recommended as a protective film for the diaper regions.

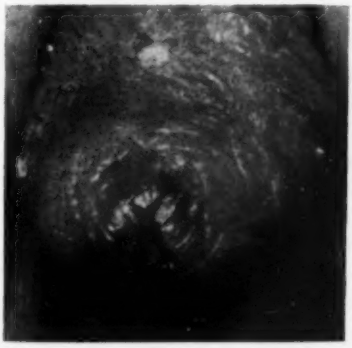

Fig. 7.—Case 7, J. L., fourth attack. Swelling of the secondarily constricted meatus. Labia showing numerous rhagades. About two and one-half times normal size.

As diet corrections, fruit juices were substituted by 50 mg. of ascorbic acid per day and skimmed milk was given. In the cases where the dermatitis described above involved only the periphery of the meatus or the glans and where the skin of the scrotum and thighs were intact, a protecting film with zinc oxide paste containing 3 per cent cod liver oil was found most successful. Herpes simplex dried up quickly after the application of zinc stearate powder as suggested by Brennemann.

## COMMENT

Ulceration and coaretation of the urethral meatus is frequently observed in clinics and private practice. Its etiology, however, is still regarded as uncertain, as the following quotations from Brennemann's textbook of pediatrics will show. Campbell, in his chapter on this subject writes: "I believe that in most instances the atresia is congenital and the important predisposing factor." Later on he states: "Moreover, I have seen cases treated by local hygiene and ointments which finally required meatotomy for cure." Brennemann, the author of this book, adds in parenthesis, "It is evident that the urologist sees the ulcerated meatus under conditions that differ from those of the pediatrician. . . . Ulceration of the meatus in circumcised infants of the diaper age is very common. It is practically restricted to artificially fed infants from 5 to 18 months of age, and it is caused by free ammonia on the night diaper. The narrowed

meatus is, here, clearly a result and not a cause of the ulceration, and in my experience has never seemed to need meatotomy."

In order to approach this problem, the urethral orifices of circumcised babies and young boys visiting the clinics were carefully checked and their anterior-posterior diameters measured. The first question that had to be answered was, What is a congenital small meatus? H. Gray,[19] in his textbook on Anatomy, states that the external urethral orifice is a vertical split bounded on either side by two small labia, its length in adults being 6 mm. The definition of a stenosis concerning this split-shaped opening is simple because it can occur only in one direction and consists of a shortening of the anterior-posterior diameter.

Careful observation of the presented cases shows that congenital shortening of this diameter can well be differentiated from secondary constriction of the orifice, which occurs due to inflammatory scarring. The lateral labia are completely intact in congenital stenosis, and the split-shaped opening is well preserved. In secondary stenosis, there are fissures or rhagades, and in many of them there is an irregularly deformed orifice.

In none of the tabled cases was the lesion seen where the anterior-posterior diameter of the urethral opening was more than 3 mm. In one of the tabled cases (20) and in three of the nonlisted children, brothers had been seen at the clinic with the same lesion. This family occurrence supports the opinion that the congenitally small meatus may be a predisposing factor of the disorder. Two patients out of the fifteen not listed, one of them 5 months, the other 8 months of age, showed a normally sized urethral orifice with an anterior-posterior diameter of more than 3 mm. In these two cases the ulceration was brought about by an enormous inflammatory swelling due to ammonia dermatitis without predisposing stenosis of the meatus. The other patients not listed had diameters measuring up to 3 mm. except for two patients with a shorter diameter required bougie dilatation and two patients with a pin-point opening who had to undergo meatotomy. If, in cases of congenital stenosis, the diameter is further shortened by swelling caused by a dermatitis, the disorder is likely to occur. In fifteen of the twenty-six tabled cases and in ten of the sixteen not listed, the meatus showed only a moderate degree of narrowing (1 to 3 mm. anterior-posterior diameter length). This series probably contains borderline cases of stenosis not definitely proved. However, it seems likely that even moderate stenosis constitutes a predisposing factor. Further investigations on a larger scale will be necessary to decide whether stenosis is an essential cause, or whether inflammation around the urethra, as seen in ammonia dermatitis, may be the only etiologic factor.

## SUMMARY

1. Superficial ulceration and coarctation of the urethral orifice has been observed in forty-one cases.

2. Its etiology has been discussed, and in all cases but two, congenital shortening of the anterior-posterior diameter of the external meatus has been found.

3. Contributing skin factors are: (a) most frequently, ammonia dermatitis, (b) a dermatitis of unknown origin affecting only glans and the periphery of the meatus, (c) herpes simplex.

4. The dangers of obstructive uropathy caused by this disorder have been stressed.

5. Treatment of the disorder has been suggested.

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# SEX PRECOCITY AND THE ADRENOGENITAL SYNDROME

BEATRICE BERGMAN, M.D.
NEW YORK, N. Y.

THE purpose of this paper is to present the present-day concepts of sex precocity and the adrenogenital syndrome, with case histories. An attempt will be made to clarify some of the confusion that exists for the pediatrician regarding these interesting problems, and to outline the current management.

The subject has excited interest from the earliest days of antiquity and references concerning such abnormal individuals can be found in the early Greek writings and in the Old Testament. An excellent historical account may be found in Young's[1] book, describing these abnormalities and recounting the widespread interest that has existed throughout the ages.

## DEFINITIONS

In order to have a common basis of understanding, the terminology used in this discussion will be defined.

1. Sex precocity is a condition of childhood characterized by genital development which approaches adult proportions during the first eight to ten years of life.

2. Precocious puberty is sex precocity with further maturation of the child, as manifested by spermatogenesis and menses.

3. The adrenogenital syndrome is a condition characterized by sex precocity resulting from a disturbance of the adrenal cortex, either due to hyperfunction, hyperplasia, or neoplasm, and often associated with metabolic disturbances, known as Cushing's phase of the syndrome.

4. Pseudohermaphroditism is a condition characterized by lack of clear-cut differentiation of the external genitalia, while the internal genitalia are isosexual. This is due to a hyperplasia of the adrenal cortex in utero, and is an instance of the adrenogenital syndrome.

5. True hermaphroditism, on the other hand, is a developmental defect in embryonic life, and is probably not related to the disturbances of the adrenal cortex. The external genitalia in this condition may be of both sexes and the internal genitalia consists of both ovaries and testes, or ovotestes.

## DISCUSSION

Sex precocity is usually divided into two etiological groups:

I. the cerebral or hypothalamic group, and

II. the endocrine or abdominal hormonal group[2]

I. *The Cerebral or Hypothalamic Group.*—

This group is due to intracranial lesions, usually situated on the floor of, or in close proximity to, the third ventricle. These lesions are believed to

From the Department of Pediatrics, New York Post-Graduate Medical School and Hospital, Columbia University.

activate the anterior lobe of the pituitary gland, particularly its gonadotropic hormone, which secondarily stimulates the gonads to secrete sex hormones. More recently, some observers[3] have expressed the opinion that there exists a sex center in the posterior hypothalamus, and that tumors in and around the third ventricle, and those of the pineal gland, act by secondary pressure on this center. Through neural or neurohumeral means, the anterior pituitary gland is activated.

CASE SUMMARY

The cerebral or hypothalamic group is exemplified by a case previously reported[4] in a female child (Fig. 1), 3 years and 2 months of age, admitted with the complaints of inability to talk, walk, and stand, or use the right arm or leg. Physical examination revealed a well-developed and nourished child showing evidence of adiposity and premature sexual development, large mature breasts, external genitalia comparable to those of a child at the age of puberty, and sparse pubic hair. A right hemiparesis, marked ankle clonus of the right foot, and bilateral optic atrophy were also noted. A ventriculogram showed a large mass filling the region of the third ventricle, with an enlarged ventricular cyst on the left and an internal hydrocephalus. A craniotomy was done, the third ventricle was entered, and the cyst was incised, cauterized, and removed. Postoperatively, the child showed signs of improvement. She was brighter and recognized people, her entire behavior pattern improved, and there was some regression of the genitalia and slight atrophy of the breasts.

Tumors of this type, including pinealomas, always produce an isosexual precocity. These tumors are evidenced by intracranial pressure, failing vision, vomiting, headache, and internal hydrocephalus. This condition progresses slowly and usually produces a true precocious puberty. The age incidence is later than those of the endocrine type. When these conditions occur in the female child, the urinary estrogens are increased. In the male, there is an increase in the urinary androgens, but not to the extent that is present in the adrenogenital syndrome. This is a helpful differential point between the two conditions. To sum up, an isosexual precocity in a child with signs of increasing intracranial pressure should lead one to suspect a tumor in or about the hypothalamic region.

*II. The Endocrine Group.—*

Before discussing the endocrine group in detail, a brief review of the recent advances in the metabolism of the steroid hormones should prove of great interest and value. The identification and evaluation of these steroid hormones and their component fractions have been of enormous help in the differentiation of the various types of sex precocity.

Normally, males and females exerete both androgenic and estrogenic compounds in the urine. The urinary androgenic-estrogenic relationship is an index of the degree of masculinity and femininity of the individual. These hormones originate both in the gonads and in the adrenal cortices. Some of the adrenal cortical and testicular hormones are converted into neutral 17 ketosteroids, and excreted in the urine. The 17 ketosteroids found in the urine are of two types, the phenolic, and the nonphenolic, or neutral fraction. The urinary products of androgen metabolism are present in the nonphenolic or neutral fraction. Callow and co-workers[5] applied the term

17 ketosteroids to those steroids with a ketone group on the 17 carbon atom and a free methylene group. These are further divided into alpha and beta fractions. The alpha neutral ketosteroids are believed to arise from the adrenals and the gonads, while the beta ketosteroids are excretory products of the adrenal cortex. The beta fraction excreted in the urine is about 10 to 15 per cent of the total urinary output, and the alpha about 85 to 90 per cent of the total neutral ketosteroids. Talbot and associates[6] estimate the total daily excretion of neutral 17 ketosteroids in children under 7 years of age as

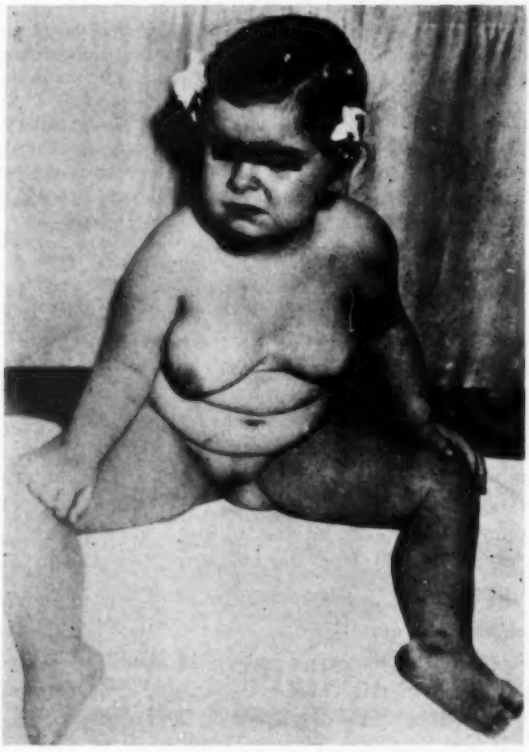


Fig. 1.—A. M. L., aged 3 years. Precocious puberty due to a cyst of the floor of the third ventricle.

0.13 mg. per twenty-four hours, between 7 to 12 years as 4 mg. per twenty-four hours, and between 12 to 15 years as 8.2 mg. per twenty-four hours. In adenomas of the adrenal cortex there is an increase in the total excretion of the neutral 17 ketosteroids and an increase in the per cent of the beta fraction. In adrenal cortical carcinoma the greatest increases occur, and findings up to 857 mg.[7] per twenty-four hours have been reported. Zimmermann[8] developed a color reaction for androgens, and this chemical test is used today, replacing the more difficult biological assay method. Steroids with a ketone group on the 17 carbon atom develop an intense absorption band with a maximum in the green.

Further fractionation of the ketosteroids into the alpha and beta fractions, has been of value in determining pathological states. According to Soffer,[9] in the urine of normal men and women these compounds have been isolated: androsterone, dehydroisoandrosterone, 3-a-hydroxy-etiocholane-17-one, pregnanediol, and estrogens. In hyperfunction, either due to hyperplasia or tumors of the adrenal cortex, additional pathological fractions isolated are: delta 3,5,-androstadiene-17-one, pregnanediol, pregnane-3,17,20-triol, and 3-a-hydroxy-androsten-17-one.

Dorfman and associates[10] isolated an androstenol-3 (alpha)-17-one, an androgen, from the urine of a girl suffering from virilism. This was identical to the compound isolated by Wolfe and co-workers[11] from the urine of a 10-year-old girl suffering from malignant adrenocortical tumor.

Not only are the total excretions of the 17 urinary ketosteroids increased in hyperfunction of the adrenal cortex, but also, certain pathological fractions have been isolated that are not present in normal urine. The highest excretion is found in adrenocortical neoplasm, as previously mentioned. Low excretion of 17 ketosteroids occurs in malnutrition, anemia, and may entirely disappear in Addison's disease.

The endocrine group includes those conditions where a hyperfunction, hyperplasia, or a neoplasm of the testicular, ovarian, or adrenal cortical cells exists. It is interesting to note that these organs involved have a common embryological origin and that they all secrete steroid hormones.

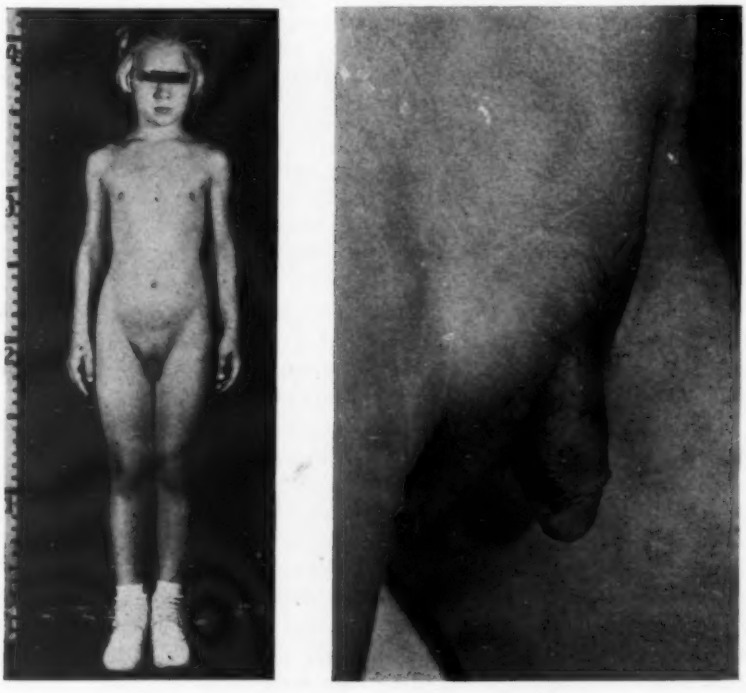
In boys, involvement of the testes causing sex precocity is usually due to a teratoid embryoma or an adenoma of the interstitial cells. These occur most commonly from the ages of 3 to 6 years. They produce an isosexual precocity. There is usually a palpable tumor mass and a slight increase of the urinary 17 ketosteroids. In girls, granulosa cell tumors of the ovary are often found, rarely teratoid embryomas or chorioepitheliomas. These produce an isosexual precocity, and may manifest themselves by either sex precocity or precocious puberty. Large amounts of estrogenic hormones are excreted in the urine. An enlargement of the abdomen is present, and a tumor mass may be palpated. Surgical removal of the mass will cause regression of the symptoms. There are other instances of sex precocity in girls for which no definite etiology can be determined, even after exhaustive study. The Albright[12] syndrome is included in the endocrine group. This is a condition in which there is sex precocity in girls, osteoporosis, and pigmentation of the skin.

## THE ADRENOGENITAL SYNDROME

In disturbances of the adrenal cortex due to hyperfunction, hyperplasia, or neoplasm, there are three age groups in which the syndrome may manifest itself: in utero, in childhood, and in adult life. There is a familial tendency to sexual abnormalities in the adrenogenital group.[13]

When the onset of the syndrome occurs in utero, pseudohermaphroditism is present without any clear-cut differentiation of the external genitalia. Determination of the sex is then difficult because of the malformations. It has been established that the majority of these cases, however, are female.

In the male, during childhood, there is precocious virilism. Sex precocity of the isosexual type is present with adult genitalia, pubic hair, deep voice, acne, increased height for age, heavy bony structure, hypertrophied musculature, and hirsutism. Hence, the term "Little Hercules" is applied to these children. Early epiphyseal closure occurs, so that while these children are initially tall for their chronological age, they develop into comparatively short men. Spermatogenesis is usually absent. The urinary 17 ketosteroids are increased, however.



*A.* *B.*

Fig. 2*A*, *B*, *C*.—*A*, L. A., aged 4½ years. Female pseudohermaphrodite. Note enlargement of clitoris and increase in height. *B*, shows marked enlargement of clitoris and pubic hair.

These children have a lack of interest in the opposite sex and generally present few psychosexual problems.[20] The prognosis is usually good in boys with the adrenogenital syndrome, since the more common etiology is hyperfunction rather than hyperplasia or neoplasm of the adrenal cortex. The condition, as a rule, resolves itself in later life so that these individuals are indistinguishable from normal adults, except that they may be more muscular and hirsute.

In the female, a heterosexual pseudoprecocity is present. There is an increase in height, hirsutism, deepening of the voice, male configuration, and no mammary development. The internal genitalia are female. The external genitalia are pseudohermaphroditic, manifested by an enlarged clitoris which resembles a hypospadic penis with the urethra opening below the clitoris. Pubic

hair appears early but is usually female in distribution. A prostate may be found. The vagina is not completely descended and often opens into the urethra. The child resembles a cryptorchid boy. Menstruation does not occur. The 17 ketosteroids are markedly increased and are in far greater excess than in the boy with the same syndrome. The prognosis for a normal sexual life is doubtful.

Fig. 2C.—Same child at 10 years of age. Note masculine configuration. Clitoris has been amputated.

An androgenic zone[14] normally exists in the fetal adrenal gland and due to some pathological condition, possibly pituitary overstimulation, it may persist throughout childhood. The masculinizing hormone is believed to originate in this androgenic zone. The hyperfunction of the adrenals that occurs in the female pseudohermaphrodite is caused by hyperplasia of the cells in this androgenic zone. Grollman[15] states that there is a marked difference in function between these cells and other portions of the adrenal cortex which control electrolyte balance, i.e., the inter-renal cells. Encroachment of the androgenic zone on the inter-renal cells might lead to Addison's disease with pigmentation, weakness, and often death.

## CASE REPORTS

CASE 1.—L. A. (Fig. 2), aged 10 years, was noted at birth to have an abnormally enlarged clitoris. At 8½ months of age, physical examination revealed a well-developed penis with glans and foreskin. There was an associated female urethra with well-developed labial folds and a palpable vagina. At 4 years, 8 months, the clitoris resembled a normal sized penis for a 9-year-old male with complete hypospadias and an urethra which opened in the normal female position. Posterior to the urethra, there was a vaginal opening. The vagina and urethra were enclosed in the labia majora. No testes were palpable, either in the labia or in the inguinal canals. X-ray of the skull revealed a sella turcica 40 per cent below average measurements. X-ray of the long bones showed them as short and stocky with an osseous and epiphyseal development of more than double that of the given chronological age. Neurologic examination was negative for signs of any pineal, pituitary, or craniopharyngeal tumors or cysts. Perirenal insufflation and cystoscopy were normal. The twenty-four hour urinary excretion of 17 ketosteroids was 32.9 mg. (normal 2 to 4 mg.).

Laparotomy revealed normal female ovaries, uterus, and Fallopian tubes. No testes were found. The suprarenal glands were normal in position but slightly enlarged. There appeared to be an adenoma of the right suprarenal. The adenoma and one-third of the left suprarenal were removed. The penile organ was amputated after ovarian tissue was demonstrated on biopsies of the ovaries. Postoperatively, the 17 urinary ketosteroids fell to 15.1 mg. per twenty-four hours. After a one-month interval, the ketosteroids rose to 24.6 mg. in twenty-four hours. Two years later, the 17 ketosteroids were 20.3 mg. in twenty-four hours. During the next two-year period, the child became more completely masculine in appearance. There was marked bony and muscular enlargement, acne of the face, deepening of the voice, no mammary development, although the pubic hair was still female in distribution. A vaginogram revealed a normal sized vagina. Psychologically, her interests were masculine. She preferred to play football and indulged in other competitive boys' sports. The urinary 17 ketosteroids, four years after the child was first seen in our clinic, had risen to 112.5 mg. in twenty-four hours.

*Case Discussion.*—This is a case of a pseudohermaphrodite that obviously originated in utero because the malformation of the genitalia was present at birth. It has been established that cases of this type show a bilateral adrenal cortical hypertrophy. Speculation arises as to whether the hypertrophy is primarily of pituitary origin,[16] since we know that there is an adrenal cortical hormone which maintains the adrenal cortex, and that the integrity of the adrenal cortex depends on this normally functioning adrenotropic hormone. If destruction of the anterior lobe of the pituitary occurs, the adrenal cortex atrophies, and death results. The child was proved to be a female pseudohermaphrodite by laparotomy, which revealed a uterus, ovaries, Fallopian tubes, and the absence of testes. The laparotomy also confirmed the hypertrophy of the adrenal cortices. The question now arises as to why this child, who was subjected to a bilateral subtotal adrenalectomy, should go on to such marked masculinization during the four-year period. The answer is perhaps indicated by the marked increase in the total urinary 17 ketosteroids in the last determination, 112.5 mg. in 24 hours. This marked rise is believed to be generally associated with an adrenocortical neoplasm. Such neoplasms may be located either in the adrenal glands proper, or in an adrenal-cortical rest situated somewhere in the abdomen between the kidneys and the pelvis.[17] The ketosteroids are being fractionated to determine if there are any pathological components present. The

child is also being subjected to further intensive workup, and tests are being repeated for location of a suspected tumor. She is being treated with estrogenic hormone at present, and an enlargement of the breasts has been noted.[18-19]

CASE 2.—L. T. (Fig. 3), aged 10 years, whose sex at birth could not be determined, was first seen in our clinic at 5 months of age. Examination at that time revealed a well-formed penis and labia. It was felt that the child was a boy with a hypospadias. No testicles were palpated. The child was given A.P.L. (anterior pituitary-like) hormone without apparent results except for an increase of penile growth and the appearance of pubic hair. Male hormones were then added to the therapy. At 4 years of age, it was decided to do plastic repair of the hypospadias, and to free the penis, which was found adherent on its undersurface. At that time the penis was 4 cm. long. Plastic reconstruction was begun, and when the child was admitted for the second stage of this repair, a tumor of the right breast was noted. In all, there were six admissions for surgical repair of what was believed to be a complete hypospadias. At 10 years of age, the child was readmitted for an exploratory operation for undescended testicles. It was noted at that time that the child's breasts were tender and enlarging, and that there was a complaint of lower abdominal pain. At operation, bilateral ovaries (confirmed by biopsy), uterus, and Fallopian tubes were found.

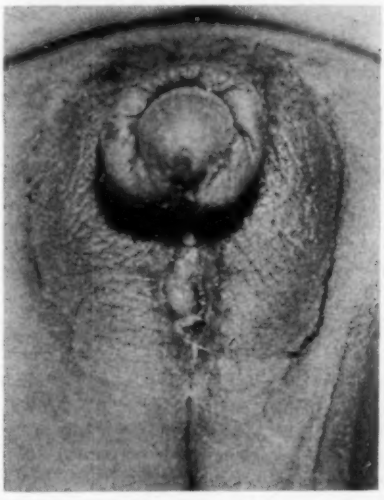


Fig. 3.—L. T., female pseudohermaphrodite aged 4, showing enlargement of clitoris and presence of labia and pubic hair

*Case Discussion.*—This is another instance of a female pseudohermaphrodite, on whom, unfortunately, the diagnosis was not made earlier because of the failure to do an exploratory laparotomy, and also because at that time the 17 ketosteroid determinations had not been developed. It is important to stress the fact that when the sex cannot be readily determined, an exploratory laparotomy must be performed. This child was reared as a boy, underwent plastic surgical repair, and was given male hormone therapy. As a result, the child has been masculinized both psychologically and endocrinologically and brought to puberty earlier than the chronological age would warrant. When the true

sex was established by laparotomy, the parents refused to consider the child a girl, and as a result the psychological management has become a serious problem.

CASE 3.—G. K. (Fig. 4) was first admitted at 11 days of age for sex determination. The genitalia revealed a small short penis with foreskin insufficient to cover the glans, and no urethral opening. The meatus was located below the penile organ and the scrotal sac was invaginated. The admitting physician reported two testicles palpated in the lower end of both inguinal canals. The diagnosis of complete hypospadias with presence of scrotal tissue was made. Two years later, the child was admitted for operation to repair the hypospadias. At that time, it was stated in the description of the genitalia that no scrotum was present, no testicles were felt, and that there was an absence of the vagina.

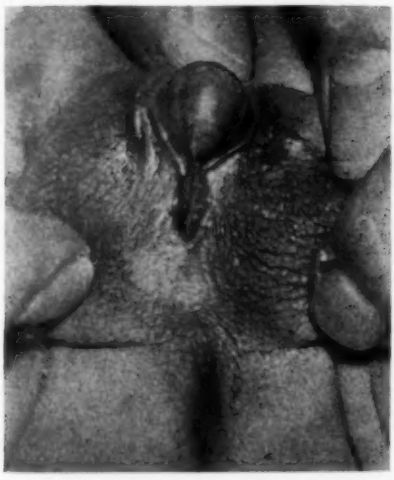


Fig. 4.—G. K., aged 2. Male pseudohermaphrodite with marked hypospadias. Note scrotal tissue.

The first stage repair for hypospadias was performed, and the diagnosis at that time was hypospadias with bilateral undescended testicles. The child was readmitted two years later for further operation to correct the hypospadias, but because there still existed some doubt as to the true sex, an exploratory laparotomy was performed. No uterus was seen, but inspection revealed the presence of what appeared to be ovaries and Fallopian tubes. Traction on these structures indicated that the tubes joined in the midline in a membranous diaphragm. The left ovary was considered to be larger than the right. Biopsy of the ovary was taken, and upon examination revealed it to be testicular tissue!

*Case Discussion.*—This child is a male pseudohermaphrodite. The clinical difficulty one encounters in diagnosing the true sex of these children is again apparent. As previously mentioned, the etiology in this case is probably due to hyperplasia of the adrenal cortex occurring in utero and stresses the importance and necessity of a complete workup before establishing the sex of the individual.

CASE 4.—E. F. (Fig. 5) was apparently normal at birth, but was noted to have enlarged genitalia at about 1 year of age. X-ray of the skull at 18 months of age, in another hospital, demonstrated it to be somewhat large in size for the stated age. There was no evidence of

intracranial tumor. The sella turcica was normal in size and shape. The diagnosis at that hospital was macrogenitosomia praecox and premature puberty. X-ray of the bones showed markedly advanced bone age. Exploration of the adrenals was done one year later, and showed no evidence of malignancy. Biopsy of the rib, testes, and epididymis was also negative for malignancy.

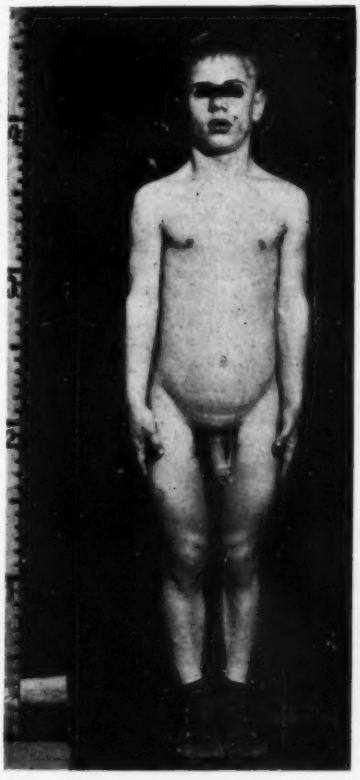


Fig. 5.—E. F., 4½ years of age. Adrenogenital syndrome in a boy with increased masculinization and adult genitalia ("Little Hercules").

He was seen at our clinic at 4½ years of age. He weighed 75½ pounds, and was 4 feet, 3¾ inches tall, which was twice the size for his age. He was well proportioned, quite muscular, with adult genitalia, but no prostatic enlargement. Pubic hair, deepening of the voice, and acne of the face were present. His mental development was superior. The basal metabolic rate was plus 8, and the urinary 17 ketosteroids were 9.2 mg. in twenty-four hours (normal for this age is less than 5 mg. per twenty-four hours). He is being followed in the clinic at present, attends school, and is in his proper grade. The school was originally fearful of admitting this child, but he is adjusting very well to the classroom situation and has shown no abnormal interest in the opposite sex.

*Case Discussion.*—It is our impression that we are dealing with a case of the adrenogenital syndrome due to hyperfunction in a boy, manifested by increased masculinization, giving the so-called "Little Hercules" type. The absence of

CHART I

| AGE AND SEX | HYPERFUNCTION, HYPERPLASIA OR NEOPLASM OF THE ADRENAL CORTEX | DIFFERENTIAL DIAGNOSIS |
|---|---|---|
| 1. In Fetal or Intrauterine Life | *Pseudohermaphroditism*<br>No clear-cut differentiation of external genitalia. The earlier the abnormality, the more marked are the physical and sexual changes. | *True hermaphroditism*<br>An embryological developmental defect, in contrast to pseudohermaphroditism and is not related to abnormalities of the adrenal cortex. |
| 2. Childhood<br>*Boy* | Manifests itself after birth: accentuation of the male characteristics; adult genitalia; pubic hair; initially tall; later, normal or small for age; early closure of the epiphysis; increase of the 17 urinary ketosteroids; isosexual precocity; lack of sexual aggressiveness in boys: condition resolves itself: cause, usually a hyperfunction without hyperplasia. | *Pineal tumors*<br>Occur in boys only, from 4 to 10 years of age: symptoms of intracranial pressure, internal hydrocephalus, failing vision, headache, vomiting; mental precocity and sexual precocity; possibly due to pressure on hypothalamic region; no proof of real pineal hormone: increase of ketosteroids.<br>*Testicular tumors*<br>Occur in boys 4 to 6 years of age; tumor of the interstitial cells; isosexual precocity: evidence of tumor mass present in testicle; increase in ketosteroids.<br>*Cerebral tumors*<br>Involving the floor of the third ventricle or hypothalamus; produce isosexual precocity or precocious puberty with increase in ketosteroids; usually slow growing; symptoms of intracranial pressure. |
| *Girl* | Heterosexual pseudoprecocity; pubic hair; enlarged clitoris; hypertrichosis; male configuration; no mammary development; marked increase in 17 neutral urinary ketosteroids; no menstruation; prostate may be found, and aberrant adrenal cortical tissue; prognosis poor for normal sexual function. | *Granulosis cell tumors of the ovary*<br>True isosexual precocity; enlargement of the breasts; pubic hair; menstruation; marked increase in urinary estrogens; tumor mass present.<br>*Cerebral tumors*<br>Early isosexual precocity; evidence of intracranial pressure; slow growing; normal urinary estrogens; enormous breast enlargement.<br>*Sex precocity*<br>May occur early in females, usually of unknown cause. Albright's syndrome, sex precocity in girls only; osteoporosis and skin pigmentation. |
| *Cushing's Syndrome* | Signs of Cushing's syndrome may occur in both sexes associated with the adrenal genital syndrome. Among the symptoms are: obesity, hypertension, glycosuria, osteoporosis, cyanosis, plethora, metabolic changes and disturbance in the carbohydrate metabolism; etiology, usually a malignant tumor of the adrenal cortex; prognosis poor. | |

psychosexual behavior difficulties is, as mentioned,[20] characteristic. The slight increase in the urinary 17 ketosteroids is also compatible with the diagnosis. This boy is at present receiving no medication, but he is being observed and retested from time to time. It is felt that this child will develop into a normal adult of relatively short stature and that this condition will be self-limited.

## SUMMARY

In summary, the following chart is presented which may help to provide a rapid method of differentiating cases of sex precocity and the adrenogenital syndrome.

## CONCLUSION

A discussion, case summaries, and a differential diagnostic table of sex precocity and the adrenogenital syndrome have been presented, with an account of 17 urinary ketosteroids. It is hoped that this will be of value in the recognition and management of these abnormal, unfortunate individuals and that the tragic mistakes made in the past may be obviated by a knowledge of the procedures employed and the newer tests utilized in the differentiation of these cases.

I am indebted to Dr. Alfred B. Amler and Miss Virginia States for their assistance in the preparation of this paper.

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# SERUM PRECIPITABLE IODINES IN RECOGNITION OF CRETINISM AND IN CONTROL OF THYROID MEDICATION

EVELYN B. MAN, PH.D., CHARLES S. CULOTTA, M.D.,
DOROTHY A. SIEGFRIED, B.S., AND CARTER STILSON, M.D.
NEW HAVEN, CONN.

IT HAS been suggested that irrevocable damage occurs in children by the time clinical symptoms of cretinism become complete. Recognition of thyroid hypofunction before all symptoms develop would permit earlier thyroid medication. Our data have been examined to learn whether or not the measurement of serum precipitable iodine is useful in the diagnosis of cretinism and in the differential diagnosis between cretinism and mongolism. Serum precipitable iodines and cholesterols have been compared as to their usefulness in the diagnosis of cretinism. Six cretin children have been studied after treatment with desiccated thyroid.

## EXPERIMENTAL SUBJECTS AND METHODS

The subjects were boys and girls 6 weeks to 16 years of age. All of them were seen on the pediatric wards or in the Pediatric Clinic. Before blood studies were made in the nine cretin children, there was no great question, clinically, as to the diagnosis, except for one more doubtful case. The clinical diagnosis was based on the usual criteria for cretinism: retarded mental and physical development; retarded bone age by x-ray; umbilical hernia; enlarged tongue; thickened skin and coarse hair; constipation, and poor feeding history. All these symptoms appeared to a high frequency in the children presented. One patient had also severe cardiac decompensation which has since cleared completely. Iodines were determined in the serum of seventy-two noncretin children, in the effort to exclude the possibility of thyroid dysfunction; in these children no true criteria of hypo- or hyperthyroidism were demonstrable. Though some had low basal metabolic rates or were obese, these signs were not considered indicative of hypothyroidism. Some children were mentally retarded. Some were suspected of hyperthyroidism because of tachycardia, unexplained fever, or failure to gain in weight. Eleven had simple, nontoxic goiters. Eight were malnourished.

Serum iodines were determined by the Riggs and Man[1] permanganate acid ashing method. It has now become a routine technique to precipitate duplicate 6 c.c. portions of serum with zinc sulfate and sodium hydroxide.[2] With the precipitated proteins, the hormonal iodine seems to be included, while the inorganic iodine remains in filtrate and washings. Some of the older determinations

From the Departments of Psychiatry and Pediatrics, Yale University School of Medicine.
Some of the data included in this paper have been offered for presentation at the meetings of the Federation of American Societies for Experimental Biology in May, 1947, in Chicago, Ill.

of serum iodine were made on serum without precipitation and are called "total iodines." Values of total iodine, due to the inclusion of inorganic iodine, are subject to positive error, which usually does not exceed one gamma per cent unless the patient has received inorganic iodine. In the charts total iodines are designated differently than precipitable iodines. In a couple of instances when total iodines were determined, insufficient blood could be obtained from the child to permit duplicate 6 c.c. aliquots of serum. In one instance only 2.5 c.c. aliquots were used. Iodines determined on small aliquots of serum are of course open to a larger experimental error than determinations on the usual quantity of serum. These values on small aliquots have not been designated in the charts because this difficulty arose so rarely. Of the eighty-one initial determinations of serum iodine, fifty-seven measurements were of precipitable iodine and twenty-four samples were total iodines.

Serum cholesterols were evaluated by a gravimetric digitonin procedure.[3, 4]

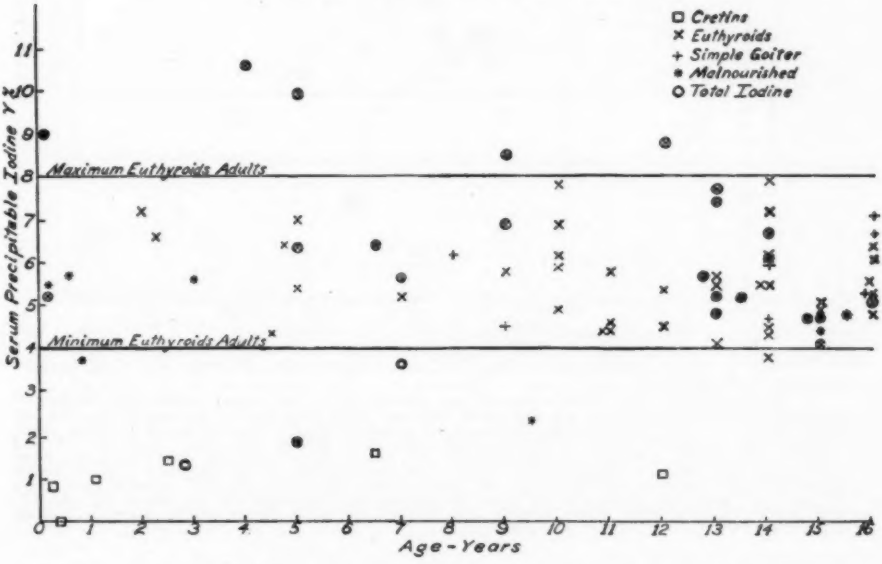


Chart 1.—Serum precipitable iodines and ages of nine cretin and seventy-two noncretin children, 6 weeks to 16 years of age.

## RESULTS

In Chart 1 are presented the ages and serum iodines of the nine cretin children and of the seventy-two noncretin children. Each point on the chart represents a different child. Except for five slightly high total iodines (the positive error of which has already been discussed) all but three of the serum precipitable iodines of the noncretin children fell within 4.0 to 8.0 gamma per cent, the range which has been defined for euthyroid adults.[5] Of the three exceptional values, two differ from normal by less than the experimental error of the method.

Only one subject, a cachectic child with anorexia nervosa, had a clearly subnormal value, 2.4 gamma per cent. Later, after gavage feeding without thyroid medication, the serum precipitable iodine rose spontaneously to 4.2 gamma per cent.

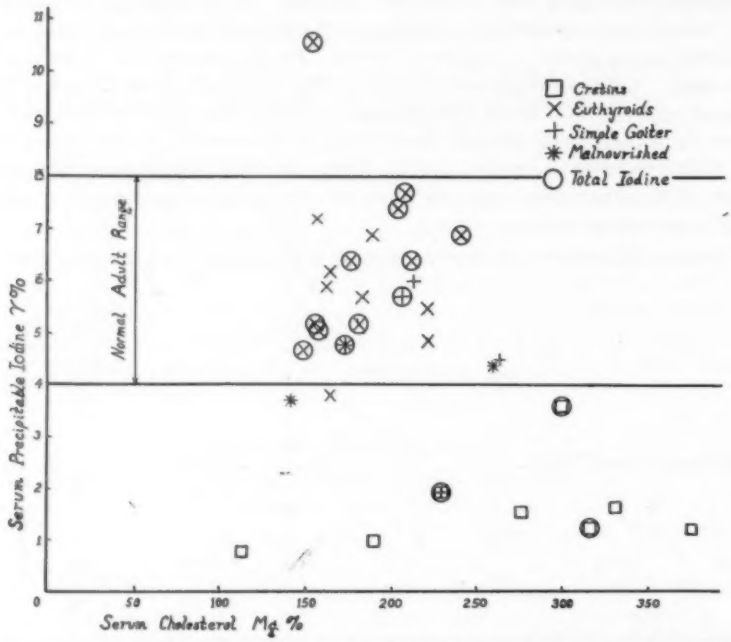


Chart 2.—Serum precipitable iodines and serum cholesterols of eight cretin and twenty-four noncretin children.

The horizontal guide lines represent the normal range for serum precipitable iodines of euthyroid adults, 4.0 to 8.0 gamma per cent.[5] Vertical guide lines for the normal range of serum cholesterol have not been drawn. In our own previous studies on euthyroid children when iodines were not determined, fifty-three boys and girls between the ages of 5 and 14 had cholesterols ranging between 114 and 296 mg. per cent.[6] Macy[7] has reported that serum lipid values may be low in young children. The cretin children ranged in age from 1 to 12 years. It would be impossible to draw any one line representing the maximum normal cholesterol for such a mixed age group. From available data[8-13] any cholesterol value above 250 mg. per cent might be considered above normal, but distinct elevation of the serum cholesterol could not be granted below 300 mg. per cent.

All values on the nine cretin children in Chart 1 were subnormal. In fact, with the exception of one less accurate total iodine, all values fell below 2.0 gamma per cent. None of these children except one had been treated with desiccated thyroid for at least four weeks prior to the measurement of serum iodine. Clinically recognizable cretinism is therefore accompanied by a distinct decrease in the serum precipitable iodine. Conversely, in many clinical conditions studied, with the exception of the one cachectic child, all low serum precipitable iodines occurred in cretin children.

In Chart 2 are shown the serum iodines and cholesterols of the children in Chart 1. The same symbols are used as in the first chart. Both iodine and

cholesterol were determined simultaneously in only eight of the cretin children and twenty-four of the noncretin children of Chart 1. Seven of the cretin children had not had desiccated thyroid for at least six weeks preceding the blood study; one of them had been on small amounts of desiccated thyroid. Of eight cretin children, three had serum cholesterols below 250 mg. per cent and only three had serum cholesterols above 300 mg. per cent. On the other hand,

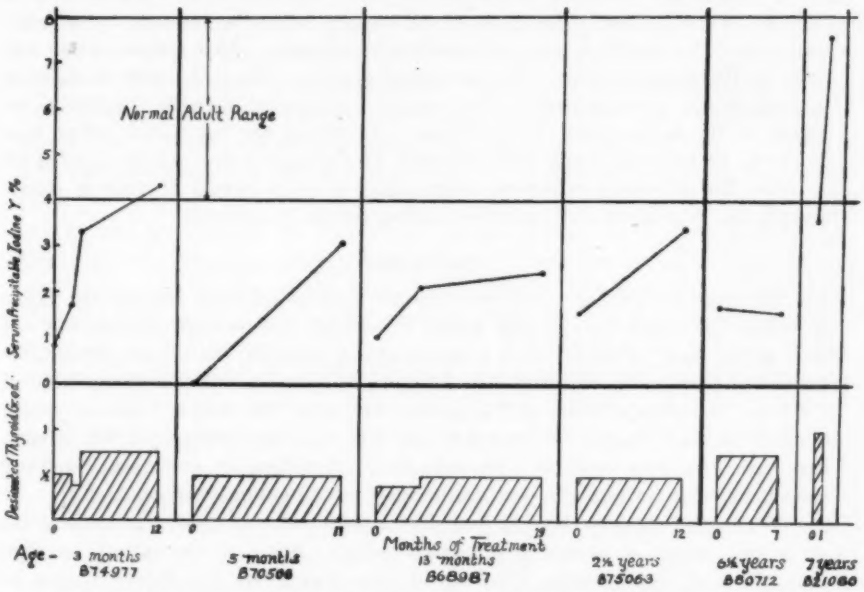


Chart 3.—Serum precipitable iodines in relation to medication with desiccated thyroid of six cretin children.

At the bottom of this chart is given the age of the child at the time of the initial iodine, when the child had not been treated with desiccated thyroid. The children are arranged in order of age when the first precipitable iodine was measured. Above the initial ages are shown the doses of desiccated thyroid per day with the duration of treatment indicated in months. Thus the total period of treatment for the first child is 12 months. No change in dosage is illustrated unless the dosage had been maintained for at least one month. When some of the children were started on desiccated thyroid, changes in the dosage were made after a few days in relation to clinical symptoms. These changes are not included. Above the thyroid dosage are given the serum precipitable iodines.

all the iodines fell at or below 2.0 gamma per cent with the exception of one total iodine. It is at once obvious that the measurement of serum cholesterol is less reliable than the estimation of serum precipitable iodine in the diagnosis of cretinism in children. It might be pointed out that the two lowest cholesterols were found in the two youngest children. The 3-month-old child had a cholesterol of 113, and the one-year-old child a cholesterol of 190 mg. per cent. Even if the serum cholesterol of infants is lower normally than that of older children, neither of these values is sufficiently elevated to aid in the diagnosis of cretinism. Therefore, while an elevated serum cholesterol is of value diagnostically, a cholesterol below 250 mg. per cent does not exclude the possibility of cretinism.

In Chart 3 are presented some of the data concerning the response of six of the cretin children, mentioned previously, to thyroid medication. Only six children have been included because no adequate follow-up work has been conducted on three. Clinically, the outstanding fact about these six children is that one had responded quite satisfactorily to administration of desiccated thyroid and two had not responded completely. These two are the third and fifth on the chart. These are the two whose serum precipitable iodines, after desiccated thyroid, were lowest. The first child on the chart is the one who, clinically, has responded in such a satisfactory manner. Her serum iodine has risen to the normal range. As was pointed out in Chart 3, brief changes in thyroid dosage were omitted. This omission prevented showing the initial response of the serum precipitable iodines. In two of the babies the iodine rose markedly at first, and then after a couple of weeks or a few months leveled off or fell. Measurement of serum iodine after a short period of time is consequently not conclusive as to whether thyroid dosage is adequate.

## DISCUSSION

The data concerning cretin children are open to criticism because the range of values for serum precipitable iodine in normal infants and children has not been established. Talbot,[14] with a micromethod, reported the serum precipitable iodines of thirty-nine children 3 to 13 years of age as varying between 4.0 and 7.0, with an average value of 5.5 gamma per cent. In eleven "normal young adults" iodines ranged between 6.0 and 8.4, with an average of 7.0 gamma per cent. In four cases of "juvenile hypothyroidism or cretinism," iodines varied between 1.8 and 3.0 with an average of 2.3 gamma per cent.

With our iodine method, at least 25 c.c. of blood is necessary at present for the measurement of serum precipitable iodine. Some of the infants studied have required transfusions after blood was drawn for the determination of serum iodine. Such heroic measures have not seemed justified as yet in order to define the range of values in normal infants. It is still a question whether the newborn infant may not have a higher normal serum iodine than the older child, since during normal pregnancy the mother has a higher serum iodine than normal nonpregnant women. It has been found in this laboratory that the average serum iodine of sixteen pregnant females was 8.4 gamma per cent, while the average for nonpregnant women was 5.6, plus or minus 1.3 gamma per cent.[5] Cord blood of two newborn infants had serum precipitable iodine contents of 7.3 and 10.0 gamma per cent.

Our values on noncretin children were collected from boys and girls in whom thyroid dysfunction was suspected. Some of their clinical symptoms were described earlier. Their iodine values may not be representative of children who develop normally. There are insufficient data to attempt any differentiation between boys and girls. The determinations on infants are too few to show whether the iodine level changes as the infant develops. Furthermore no single child has been followed during growth. With three exceptions, all the serum precipitable iodines of the seventy-two noncretin children fell within the same range as for euthyroid adults, 4.0 to 8.0 gamma per cent.

A few odd facts about the family histories of these cretin children might be mentioned. In the case of a baby (B68987) after cretinism was recognized the serum iodines of both the mother and father were measured. The serum precipitable iodines of both the mother and father were normal, 6.7 and 6.3 gamma per cent, respectively. In the case of another baby (B75063) it was learned that the mother was pregnant. During this subsequent pregnancy her precipitable iodine conformed entirely with values for normal pregnant females 9.1 gamma per cent.[5] The baby born at the end of this pregnancy and another sibling have no clinical signs of diminished thyroid function. On the other hand, the mother of one of the cretin boys (B21080) had taken desiccated thyroid during her pregnancy. This fact has little significance. Occasionally pregnant women are given some thyroid. In another research project it was learned that two women, who were studied while taking 3 grains of thyroid daily during pregnancy, have had babies who are developing normally.

It has been found in regulating thyroid dosage for hypothyroid adults that patients are both subjectively and objectively better when their serum precipitable iodines are within the normal range.[15] It was noted that the child with the best clinical response to desiccated thyroid had a precipitable iodine above 4.0 gamma per cent and that the two children with the poorest clinical responses to thyroid had iodines of 1.6 and 2.4 gamma per cent. These facts prompt the question whether control of thyroid dosage in children might be facilitated by noting whether the serum precipitable iodine has risen to the normal range.

Another query concerning thyroid dosage is illustrated in Chart 3. The first baby, who was only 3 months old, has responded satisfactorily to ¾ grain of thyroid daily. The next three children, all of whom were older, 5, 13, and 30 months respectively, were given less thyroid, only ½ grain daily. It seems possible that cretin children would be benefited by slightly larger doses of thyroid than were used, or that the dose be increased as the baby grows older. In this connection, it might be pointed out that ¾ grain daily has not seemed clinically adequate for the 6½-year-old patient, the next to the last subject on the chart. When the last child, a boy of 7 years, was on one grain of thyroid a day, thyroid overdosage was suspected because of increased irritability; the dose was diminished slightly (to ¾ grain per day). The marked increase in his iodine may be exaggerated because a total, not a precipitable iodine was determined. The rise may also be more pronounced because only one month had elapsed after initiation of one grain of thyroid daily.

Further investigation should answer two questions. Is the measurement of serum precipitable iodine an aid in the diagnosis of thyroid hypofunction before clinical cretinism can be recognized? Does a more complete clinical response occur when thyroid dosage is regulated to maintain a normal serum iodine, than when thyroid dosage is insufficient to elevate the serum iodine to 4.0 gamma per cent? It is to be recognized that while there may be a good response in physical development to medication, mental development may lag behind. This

has been tentatively ascribed to a possible irreversible central nervous system defect,[16] but it may be that this development can be furthered by the use of serum precipitable iodine determinations in the control of medication.

## CONCLUSIONS

Serum precipitable iodines were below normal adult values usually less than 2.0 gamma per cent in nine cretin children ranging from 3 months to 12 years of age. Only one of seventy-two noncretin children, a patient with anorexia nervosa, had a serum precipitable iodine distinctly below 4.0 gamma per cent.

Measurement of serum precipitable iodine is more reliable than evaluation of serum cholesterol in the diagnosis of cretinism.

The clinical response in relation to the level of serum precipitable iodine after administration of different doses of desiccated thyroid has been discussed.

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# THE DETERMINATION OF STATUS AND PROGRESS IN CHILDREN WITH NUTRITIVE FAILURE

ARVIN W. MANN, D.D.S., SAMUEL DREIZEN, D.D.S., AND TOM D. SPIES, M.D.
BIRMINGHAM, ALA.
WITH THE TECHNICAL ASSISTANCE OF S. IDELL PYLE, M.S.
CLEVELAND, OHIO

THE effect of nutritive failure* on growth† has been studied intensively at the Nutrition Clinic of the Hillman Hospital, Birmingham, Ala., since 1936. This study was prompted by the observation that many of the children accompanying parents to the clinic showed the same signs and symptoms of nutritional deficiency disease as those observed in their parents.[2] An analysis of the kind and amount of food that the family ate revealed that few if any of the children could be getting the amounts of nutriment essential for normal growth.[3] This was substantiated by clinical observations which found the children to be thin, apathetic, and small for their age. The frequent presence of the classic signs of nutritional deficiency disease in the family pets,[4, 5] which lived on scraps from the family table, provided further proof of the inadequacy of the food available for these children. The foregoing observations, together with the evidence obtained from economic histories which revealed the impoverished state of these family groups, were taken as an indication that the failure to grow in this group of children was mainly nutritional in origin.

To determine the extent to which the growth and the physical condition of these children was influenced by nutritive failure, it was decided to compare them with a group of children free from nutritional deficiency diseases. This paper has been confined to a comparison of the findings obtained when the Wetzel Grid‡ for assessing physical fitness was used in conjunction with regular medical and dietary examinations over a two-year period of time.[6]

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University of Cincinnati Studies in Nutrition at the Hillman Hospital, Birmingham, Ala. From the Department of Internal Medicine, University of Cincinnati.

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*"The term 'nutritive failure' perhaps means different things to different physicians. It is used in our clinic to describe a variable clinical picture and does not have a specific physiological connotation. It does not indicate why the nutrition has failed but simply that it has failed.

"Our work has convinced us that many times the physician cannot determine at once the physiologic basis of the nutritional insufficiency, and by the use of this term he can describe briefly the clinical picture without knowing the precise causation. The term has become so commonly entrenched in our clinical usage that it does not seem wise for us to restrict it or to change it.

"For the sake of clarity, the various disturbances and mechanisms which create a picture of nutritive failure we often describe separately. We can see pellagrous dermatitis or glossitis, beriberi neuritis or beriberi heart disease, scorbutic hemorrhages or gingivitis, cheilosis or the ocular symptoms of riboflavin deficiency as present symptoms arising as a response of the body to a long-continued deprivation of certain nutrients in food. Hence, nutritive failure is a more inclusive term than pellagra, beriberi, scurvy, or riboflavin deficiency in that it connotes varying degrees of mixed deficiencies operating simultaneously."[1]

†*Growth* is the progressive development of an organism from its earliest stages, characterized by increase in size and cell complexity.

‡The Wetzel Grid is but one of several methods which have been and are being used in the Nutrition Clinic to measure the effect of prolonged nutritive failure on the growth of children. Serial roentgenograms of the hand and forearm, photographs in the nude, and cephalometric x-rays are also being utilized, and these results will be reported separately in subsequent communications.

### SELECTION OF PATIENTS

Ninety-four children (forty-seven test patients and forty-seven control patients) were selected for this study on the basis of their nutritive status, freedom from acute and clinically apparent infectious disease, and economic background. The test group was composed of twenty boys and twenty-seven girls, ranging in age from 5 to 15 years, each of whom had a history of previous or existing nutritional deficiency disease. These children came for the most part from rural families whose incomes were insufficient to allow an adequate diet most or all of the time. By contrast, the control group of twenty boys and twenty-seven girls, ranging in age from 5 to 14½ years, were all free from nutritional deficiency diseases at the time of their selection. The children in this group came from urban families whose incomes were high enough to supply adequate amounts of dietary essentials at all times. The ninety-four children were all natives of north central Alabama, a region where nutritional deficiency diseases are endemic.

### METHOD OF STUDY

All children were brought to the clinic for a medical examination at least four times each calendar year. In most instances, many more examinations were made, especially during the spring and summer months. None of the children received vitamin, mineral, or supplemental dietary therapy at any time during the two years. A record of all manifestations of nutritive failure and acute infectious disease was kept for each child. The chronological age, height, and weight of each child was recorded at each visit, and these data were plotted on the Wetzel Grid. The nutritive value of the diet was estimated once each year from diet records of food intake for one week. These diet histories were all taken in so far as possible at the same time of year.

### OBSERVATIONS

The findings of the physical examinations appear in Table I. In the test group every child except one showed some signs or symptoms suggesting nutritional deficiency disease during the two-year period of study. In the control group only one child, a boy with photophobia and glossitis, had evidence of nutritional deficiency disease. The control group, however, appeared to be more susceptible to acute infectious diseases (probably because of their urban environment), since ten girls and thirteen boys were affected as compared with only two boys and six girls in the test group. Allergic responses to particular foods were reported for four children in the test group and three children in the control group. Asthma became acute in one child in the test group and two children in the control group during the period of the study, and one child in the test group developed hay fever. Five of the children in the test group passed worms of undefined types at one time or another during the two years. No similar condition was reported by any children in the control group.

When the dietary information was assessed,* it was noted that a definite and easily discernible difference existed between the diets of the two groups in re-

*Recommendations of National Research Council.

TABLE I. MEDICAL REPORT OF FORTY-SEVEN TEST CHILDREN AND FORTY-SEVEN CONTROL CHILDREN

| ILLNESS OR SYMPTOM | TEST CASES | | | CONTROL CASES | | |
|---|---|---|---|---|---|---|
| | BOYS | GIRLS | TOTAL | BOYS | GIRLS | TOTAL |
| *Signs and Symptoms Suggestive of Nutritional Deficiency* | | | | | | |
| Sense organs: | | | | | | |
| Ocular injection | 13 | 20 | 33 | 0 | 0 | 0 |
| Photophobia | 7 | 9 | 16 | 1 | 0 | 1 |
| Night blindness | 5 | 7 | 12 | 0 | 0 | 0 |
| Burning and itching of eyes | 10 | 10 | 20 | 0 | 0 | 0 |
| Digestive system: | | | | | | |
| Slick tongue (atrophied lingual papillae) | 6 | 6 | 12 | 0 | 0 | 0 |
| Hypertrophied lingual papillae | 8 | 9 | 17 | 0 | 0 | 0 |
| Glossodynia | 0 | 4 | 4 | 0 | 0 | 0 |
| Engorged swollen hypertrophied gingivae | 7 | 3 | 10 | 0 | 0 | 0 |
| Bilateral cheilosis | 9 | 10 | 19 | 0 | 0 | 0 |
| Riboflavin glossitis | 8 | 10 | 18 | 1 | 0 | 1 |
| Anorexia | 8 | 12 | 20 | 0 | 0 | 0 |
| Diarrhea | 3 | 3 | 6 | 0 | 0 | 0 |
| Neuromuscular system: | | | | | | |
| Calf tenderness | 1 | 5 | 6 | 0 | 0 | 0 |
| Pseudohallucinations | 3 | 7 | 10 | 0 | 0 | 0 |
| Burning sensation of extremities | 3 | 3 | 6 | 0 | 0 | 0 |
| Irritability, fear, apprehension | 3 | 8 | 11 | 0 | 0 | 0 |
| Skeletal system: | | | | | | |
| Rachitic rosary | 0 | 1 | 1 | 0 | 0 | 0 |
| Epithelial system: | | | | | | |
| Hyperkeratosis of hair follicles | 3 | 4 | 7 | 0 | 0 | 0 |
| Skin furuncles | 5 | 12 | 17 | 0 | 0 | 0 |
| General appearance: | | | | | | |
| Emaciation | 2 | 3 | 5 | 0 | 0 | 0 |
| Total | 104 | 146 | 250 | 2 | 0 | 2 |
| *Acute Infectious Diseases* | | | | | | |
| Rubeola | 0 | 0 | 0 | 3 | 0 | 3 |
| Endemic parotitis | 0 | 1 | 1 | 2 | 0 | 2 |
| Tonsillitis | 3 | 1 | 4 | 1 | 0 | 1 |
| Pneumonia | 0 | 0 | 0 | 1 | 1 | 2 |
| Septic sore throat | 0 | 0 | 0 | 0 | 2 | 2 |
| Appendicitis | 0 | 1 | 1 | 1 | 0 | 1 |
| Varicella | 0 | 3 | 3 | 1 | 0 | 1 |
| Influenza | 0 | 1 | 1 | 3 | 4 | 7 |
| Scarlatina | 0 | 0 | 0 | 0 | 1 | 1 |
| Acute otitis | 0 | 2 | 2 | 1 | 2 | 3 |
| Total | 3 | 9 | 12 | 13 | 10 | 23 |
| *Miscellaneous Diseases* | | | | | | |
| Food allergies | 0 | 4 | 4 | 3 | 0 | 3 |
| Asthma | 0 | 1 | 1 | 1 | 1 | 2 |
| Hay fever | 0 | 1 | 1 | 0 | 0 | 0 |
| Total | 0 | 6 | 6 | 4 | 1 | 5 |

spect to their nutritional value (Table II). While only two of the children in the test group were on diets which were regarded as adequate in all respects, forty-one of the forty-seven children in the control group had diets which met all the requirements of adequacy at the time they were evaluated.

TABLE II. NUTRITIONAL INADEQUACY OF THE DIETS OF FORTY-SEVEN TEST AND FORTY-SEVEN CONTROL CHILDREN

| | TEST GROUP | | | | | | CONTROL GROUP | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | GIRLS | | BOYS | | TOTAL | | GIRLS | | BOYS | | TOTAL | |
| NUTRIENT | B* | D† | B | D | B | D | B | D | B | D | B‡ | D |
| Protein | 8 | 13 | 4 | 12 | 12 | 25 | 1 | 1 | 0 | 0 | 1 | 1 |
| Riboflavin | 4 | 12 | 2 | 14 | 6 | 26 | 2 | 0 | 0 | 0 | 2 | 0 |
| Thiamin | 5 | 4 | 3 | 2 | 8 | 6 | 5 | 0 | 0 | 0 | 5 | 0 |
| Niacin | 4 | 9 | 3 | 9 | 7 | 18 | 2 | 0 | 0 | 0 | 2 | 0 |
| Calcium | 2 | 10 | 4 | 11 | 6 | 21 | 2 | 0 | 0 | 0 | 2 | 0 |
| Phosphorus | 2 | 9 | 2 | 7 | 4 | 16 | 1 | 0 | 0 | 0 | 1 | 0 |
| Iron | 6 | 8 | 4 | 5 | 10 | 13 | 3 | 0 | 1 | 0 | 4 | 0 |
| Vitamin C | 5 | 5 | 2 | 8 | 7 | 13 | 1 | 0 | 0 | 0 | 1 | 0 |
| Vitamin A | 3 | 6 | 3 | 6 | 6 | 12 | 1 | 0 | 0 | 0 | 1 | 0 |

*B=Borderline adequacy—values which hovered about the recommended allowance.
†D=Deficient—values below recommended allowance.
‡The borderline deficiencies were distributed among five children in the control group.

Figs. 1, *A*, *B*, and *C* compare graphically the different physique channel positions reached by the subjects at one time or another during the period of study. It was noted that the peak of the distribution of physique channels attained by the children in the control group centered in both the $A_1$ and *M* channels, while that attained by the children in the test group appeared in the $B_1$ channel. On the basis of sex, the peak of distribution for each group appeared as follows: boys in the control group, $A_1$ channel; boys in the test group, *M* channel; girls in the control group, $B_1$ channel; girls in the test group, $B_2$ channel. In general, therefore, the children in the control group appeared in the physique channel on the Wetzel Grid immediately to the left of that of the corresponding test group.

Table III reveals the tendency of the majority of children in both groups to change physique channels frequently during the course of study. The children in the test group averaged 2.3 different physique channels and 3.6 total physique channels per child in contrast to 2.0 different physique channels and 2.5 total physique channels per child in the control group. The children in the test group appeared to have more erratic growth progress than those in the control group. Although this did not of itself answer the question as to

TABLE III. COMPARISON OF THE NUMBER OF DIFFERENT PHYSIQUE CHANNELS* (WETZEL) AND THE NUMBER OF TOTAL PHYSIQUE CHANNELS† (WETZEL) FOLLOWED BY FORTY-SEVEN TEST CHILDREN AND FORTY-SEVEN CONTROL CHILDREN IN A TWO-YEAR PERIOD

| | NO. DIFFERENT PHYSIQUE CHANNELS | | | | NO. TOTAL PHYSIQUE CHANNELS | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| GROUP | 1 | 2 | 3 | 4 | 1 | 2 | 3 | 4 | 5 | 6 |
| Test boys | 0 | 12 | 7 | 1 | 0 | 3 | 6 | 4 | 6 | 1 |
| Test girls | 3 | 17 | 7 | 0 | 3 | 8 | 4 | 5 | 5 | 2 |
| Total | 3 | 29 | 14 | 1 | 3 | 11 | 10 | 9 | 11 | 3 |
| Control boys | 5 | 10 | 3 | 2 | 5 | 5 | 6 | 3 | 1 | 0 |
| Control girls | 6 | 19 | 2 | 0 | 6 | 10 | 6 | 4 | 1 | 0 |
| Total | 11 | 29 | 5 | 2 | 11 | 15 | 12 | 7 | 2 | 0 |

*"Different" refers to the number of dissimilar physique channels in which the child appeared during the test period; e.g., if a child passed from $A_1$ to *M* and back to $A_1$, the total would be two different physique channels.

†"Total" refers to the number of channels in which the child appeared during the test period, taking into consideration the possibility of his appearing in a given channel more than one time; e.g., if he passed from $A_1$ to *M* and back to $A_1$ the total would be three physique channels.

TABLE IV. CHANNEL READINGS IN FORTY-SEVEN TEST CHILDREN AND FORTY-SEVEN CONTROL CHILDREN AT BEGINNING AND END OF TWO-YEAR STUDY

| CHANNEL READING | | TEST GROUP | | | CONTROL GROUP | | |
|---|---|---|---|---|---|---|---|
| START | END | BOYS | GIRLS | TOTAL | BOYS | GIRLS | TOTAL |
| *Movement in Most Favorable Physique Channels ($A_1$ M $B_1$)* | | | | | | | |
| M | $A_1$ | 1 | 0 | 1 | 1 | 1 | 2 |
| $B_1$ | M | 0 | 0 | 0 | 1 | 2 | 3 |
| $A_1$ | $A_1$ | 1 | 0 | 1 | 2 | 2 | 4 |
| $B_1$ | $B_1$ | 2 | 3 | 5 | 1 | 6 | 7 |
| M | M | 2 | 0 | 2 | 4 | 4 | 8 |
| $A_1$ | M | 2 | 3 | 5 | 3 | 0 | 3 |
| $A_1$ | $B_1$ | 1 | 2 | 3 | 1 | 0 | 1 |
| M | $B_1$ | 7 | 4 | 11 | 0 | 0 | 0 |
| Total | | 16 | 12 | 28 | 13 | 15 | 28 |
| *Movement Toward More Favorable Physique Channels (Toward $A_1$ M $B_1$)* | | | | | | | |
| $B_2$ | $B_1$ | 0 | 3 | 3 | 0 | 2 | 2 |
| $A_2$ | $A_1$ | 1 | 0 | 1 | 1 | 1 | 2 |
| $A_2$ | M | 1 | 0 | 1 | 1 | 1 | 2 |
| Total | | 2 | 3 | 5 | 2 | 4 | 6 |
| *Movement Toward Less Favorable Physique Channels (Toward $A_2$-$A_4$ $B_2$-$B_4$)* | | | | | | | |
| $A_1$ | $A_2$ | 0 | 0 | 0 | 0 | 1 | 1 |
| $A_1$ | $A_2$ | 0 | 0 | 0 | 1 | 1 | 2 |
| M | $B_2$ | 0 | 1 | 1 | 0 | 0 | 0 |
| $B_1$ | $B_2$ | 0 | 4 | 4 | 0 | 1 | 1 |
| Total | | 0 | 5 | 5 | 1 | 3 | 4 |
| *Movement in Least Favorable Physique Channels ($A_2$-$A_4$ $B_2$-$B_4$)* | | | | | | | |
| $A_3$ | $A_4$ | 0 | 0 | 0 | 0 | 1 | 1 |
| $B_3$ | $B_3$ | 0 | 1 | 1 | 0 | 0 | 0 |
| $A_2$ | $A_2$ | 0 | 0 | 0 | 2 | 0 | 2 |
| $B_2$ | $B_2$ | 1 | 2 | 3 | 1 | 2 | 3 |
| $B_3$ | $B_3$ | 0 | 2 | 2 | 1 | 0 | 1 |
| $B_4$ | $B_4$ | 0 | 1 | 1 | 0 | 0 | 0 |
| $A_3$ | $A_2$ | 1 | 0 | 1 | 0 | 0 | 0 |
| $B_2$ | $B_3$ | 0 | 1 | 1 | 0 | 2 | 2 |
| Total | | 2 | 7 | 9 | 4 | 5 | 9 |

whether the cause of the physique differences was nutritional or genetic, it would seem that the nutritional influence could be responsible for the greater frequency of channel variation in the test group.

A summary of the physique channel changes occurring on the Wetzel Grid, when the children were grouped according to sex and experimental status, is shown in Table IV. These changes are recorded as the difference in the channel positions occupied by the children at the beginning and at the end of the study. Twenty-eight of the children in the test group moved about but remained in the most favorable physique channels ($A_1$ $M$ $B_1$), nine remained in the least favorable channels ($A_2$-$A_4$ $B_2$-$B_4$), five shifted into more favorable channels, and five moved into less favorable channels during the two-year period. By contrast, twenty-eight children in the control group moved about but remained in the most favorable physique channels, nine remained in the least favorable physique channels, six shifted into more favorable channels, and four moved into less favorable channels during the same period of time. It would appear that while changes in channel position, when analyzed in this way, did not offer a point of significant distinction between the two groups of children, the direction of the movement

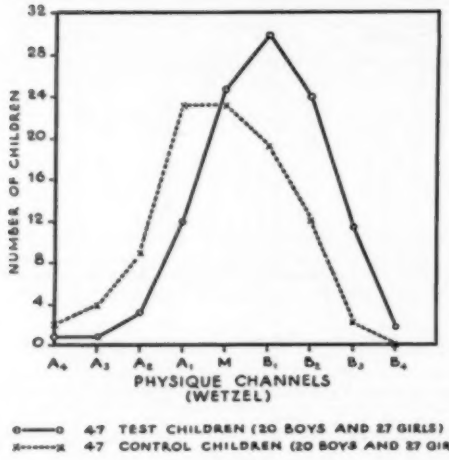


Fig. 1*A*.

Fig. 1*A*.—Graph showing the differences in the physique channels followed by the children in the test group and those in the control group during the two-year period. The children in the control group centered within the $A_1$ and $M$ channels, and the children in the test group centered within the $B_1$ channel.

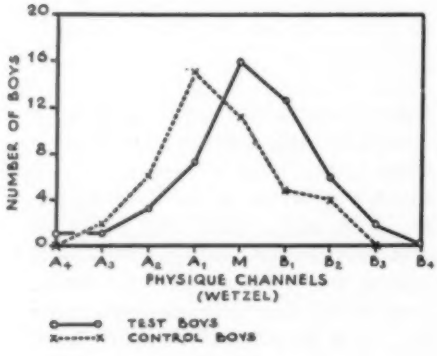


Fig. 1*B*.

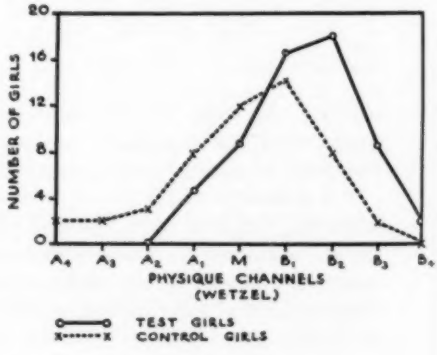


Fig. 1*C*.

Fig. 1*B*.—Graph showing the comparison of the physique channels followed by the boys in the test and control groups during the two-year period. The boys in the control group centered within the $A_1$ channel, and those in the test group centered within the $M$ channel.

Fig. 1*C*.—Graph showing the comparison of the physique channels followed by the girls in the test and control groups during the two-year period. The girls in the control group centered within the $B_1$ channel, and those in the test group centered within the $B_2$ channel.

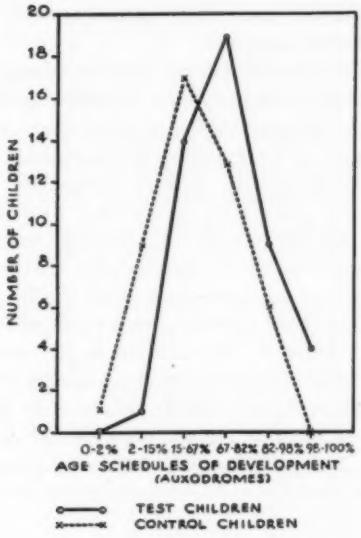


Fig. 2*A*.

Fig. 2*A*.—Graph showing the comparison of the age schedules of development (auxodromes) for children in the test and control groups. The children in the control group had for the most part faster schedules of development than did those in the test group.

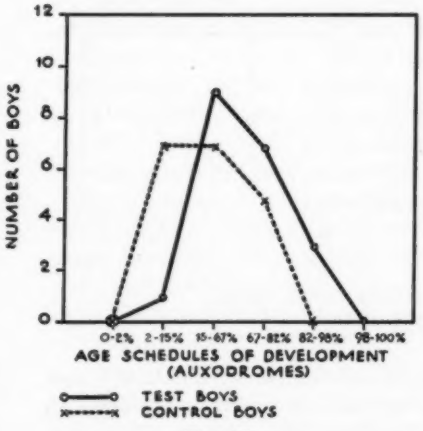


Fig. 2*B*.

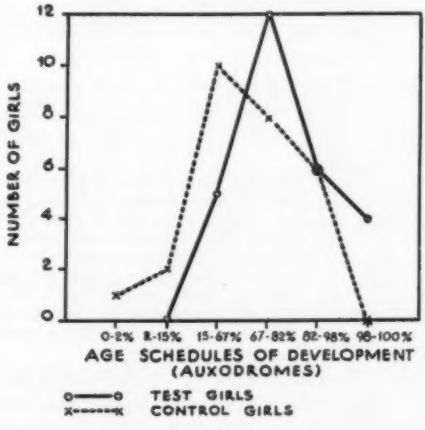


Fig. 2*C*.

Fig. 2*B*.—Graph showing that the boys in the control group had faster schedules of development than did those in the test group.

Fig. 2*C*.—Graph showing that the girls in the control group had faster schedules of development than did those in the test group.

manifested by twenty-eight children in the test group would appear to have some significance, since, in the main, they shifted toward the less favorable part of the optimal physical status grouping.

The evaluation of the changes in the physical status of the two groups of children during the period of investigation is shown in Table V. In the Grid technique, "evaluation of physical status involves the study, measurement, and interpretation of physique or body build, developmental level, and nutritional grade."[7] Taking all three factors into consideration, it was observed that twenty-six children in the test group underwent what would appear to be a detrimental change of physical status, four showed improvement, and the remainder tended to be unchanged. It would seem that in the latter group advantageous changes in one component were offset by deleterious changes in another, with the result that there was no apparent effect on the general physical status of the child. By contrast, ten children in the control group underwent a detrimental change, eight showed improvement, and the remainder were unchanged. The combination of all three factors into a single expression of physical status tended to obscure the tortuous nature of the channel progress made by the test cases. It is not clear why the ten children in the control group failed, but the evidence points toward infection rather than to primary nutritive failure.

TABLE V. COMPARISON OF THE CHANGES IN PHYSICAL STATUS OF FORTY-SEVEN TEST CHILDREN AND FORTY-SEVEN CONTROL CHILDREN DURING A TWO-YEAR PERIOD

| PHYSICAL STATUS | | | TEST CASES | | | CONTROL CASES | | |
|---|---|---|---|---|---|---|---|---|
| BODY BUILD* | DEVELOPMENTAL LEVEL† | NUTRITIONAL GRADE‡ | BOYS | GIRLS | TOTAL | BOYS | GIRLS | TOTAL |
| → | − | ↘ | 11 | 15 | 26 | 5 | 5 | 10 |
| → | + | ↘ | 2 | 0 | 2 | 2 | 0 | 2 |
| ↔ | + | ↑ | 3 | 5 | 8 | 4 | 5 | 9 |
| ↔ | 0 | ↑ | 0 | 0 | 0 | 1 | 0 | 1 |
| ↔ | − | ↑ | 3 | 3 | 6 | 5 | 9 | 14 |
| ← | + | ↖ | 1 | 3 | 4 | 3 | 5 | 8 |
| ← | − | ↖ | 0 | 1 | 1 | 0 | 3 | 3 |

*→ Moved toward *B* side of Grid during study.
↔ Was in same channel at end of study as at beginning.
← Moved toward *A* side of Grid during study.
†− Gained less than one level per month.
0 Gained one level per month.
\+ Gained more than one level per month.
‡↘ Undernutrition, indicated by a slope less than the channel system.
↑ Optimal nutrition, indicated by an up-channel advance.
↖ Overnutrition, indicated by a slope greater than the channel system.

The character of the physical progress as ascertained by the child's own age schedule of development is compared for the two groups of children in Figs. 2, *A*, *B*, and *C*. The peak of the distribution of age schedules of development for the children in the control group appeared in the 15 to 67 per cent auxodrome, while that of the children in the test group appeared in the 67 to 82 per cent auxodrome. On the basis of sex, the peak of distribution for each group appeared as follows: boys in the control group, 2 to 67 per cent auxodrome; boys in the test group, 15 to 67 per cent auxodrome; girls in the control group,

15 to 67 per cent auxodrome; and girls in the test group, 67 to 82 per cent auxodrome. In general, the children in the test group had a slower age schedule of development than those in the control group. For the most part, the children in the test group were below, and the children in the control group were above the 67 percentile auxodrome, which Wetzel regards as the medical normal.

The changes in the age schedules of development are recorded in Table VI. Thirty-six of the children in the test group and twenty-three of those in the control group were retarded, as compared with eleven of the children in the test group and twenty-four of those in the control group who were unchanged or who improved their schedule of development during the two-year period.

TABLE VI. COMPARISON OF THE CHANGES IN AGE SCHEDULES OF DEVELOPMENT (AUXODROMES) IN FORTY-SEVEN TEST AND FORTY-SEVEN CONTROL CHILDREN IN A TWO-YEAR PERIOD

| | AGE SCHEDULE OF DEVELOPMENT | | | | | | TOTAL AUXODROME CHANGES | | |
|---|---|---|---|---|---|---|---|---|---|
| | ABOVE 67 PER CENT AUXODROME | | | BELOW 67 PER CENT AUXODROME | | | | | |
| GROUP | ADVANCED | UNCHANGED | RETARDED | ADVANCED | UNCHANGED | RETARDED | ADVANCED | UNCHANGED | RETARDED |
| Test boys | 1 | 2 | 8 | 2 | 2 | 5 | 3 | 4 | 13 |
| Test girls | 0 | 0 | 6 | 2 | 2 | 17 | 2 | 2 | 23 |
| Total | 1 | 2 | 14 | 4 | 4 | 22 | 5 | 6 | 36 |
| Control boys | 5 | 3 | 8 | 1 | 2 | 1 | 6 | 5 | 9 |
| Control girls | 4 | 6 | 6 | 1 | 2 | 8 | 5 | 8 | 14 |
| Total | 9 | 9 | 14 | 2 | 4 | 9 | 11 | 13 | 23 |

When divided on the basis of their previous schedules of development, eighteen children in the control group who were above the 67 per cent auxodrome either picked up speed or remained unchanged in their schedule, while fourteen slowed down. Six of the fifteen controls who were on a schedule below the 67 per cent auxodrome advanced or remained unchanged in their growth progress, while nine slowed down. Case records indicated that infection played a large part in the retardation observed in this group of children.

By contrast, three children in the test group who were above the 67 per cent auxodrome advanced or remained unchanged in age schedule of development, while fourteen were retarded. Eight of the thirty children in the test group who were on a schedule below the 67 per cent auxodrome advanced or remained unchanged, while twenty-two slowed down. It would appear that in this group malnutrition was concurrent with the decrease in their schedules of development.

The findings reported in Table VII show the number of children who had simultaneous losses in both channel and schedule of development on the Grid throughout the two-year period, a criterion suggestive of nutritional interference with growth.[8, 9] Eleven of the children in the control group and eighteen in the test group underwent auxodromic slowing down together with an undesirable change in the physique channel. Again, infectious disease appeared to have played a major role in the changes observed in the children in the control group. While many of the children in the test group were decelerated

TABLE VII. COMPARISON OF SIMULTANEOUS CHANNEL-AUXODROME CHANGES IN FORTY-SEVEN TEST CHILDREN AND FORTY-SEVEN CONTROL CHILDREN IN A TWO-YEAR PERIOD

| SIMULTANEOUS CHANNEL-AUXODROME CHANGES | TEST GROUP | | | CONTROL GROUP | | |
|---|---|---|---|---|---|---|
| | BOYS | GIRLS | TOTAL | BOYS | GIRLS | TOTAL |
| Improved | 0 | 1 | 1 | 3 | 2 | 5 |
| Unchanged | 14 | 14 | 28 | 12 | 19 | 31 |
| Impaired | 6 | 12 | 18 | 5 | 6 | 11 |

during the period of observation, their channel changes were such that they oscillated fairly rhythmically within a limited channel range, a factor which tended to obscure the results when measured in this manner.

## DISCUSSION

In the use of height-weight determinations as indicators of growth progress of children, the tendency has been to compare the values obtained for the individuals with the mean or mode of a group heterogeneous in physique. This method of measurement obscured the principle that the interplay of genetic and environmental forces individualizes rather than generalizes. The Wetzel Grid technique, in which each child is his own standard of comparison, offers a method whereby height-weight determinations may be analyzed on an individual basis. The result is that variations of environmental origin which affect the growth process can be interpreted with a greater degree of individual accuracy. Therefore, to understand the effects of nutritive failure on growth, the patients used in this study were selected with the purpose of reducing the number of diverse genetic and environmental influences to a minimum. Genetically, both groups had a common background in that they were of native American stock whose families had lived in this region for generations. From the environmental standpoint, the groups were similar in that they were both free from acute infectious disease at the time of their selection. They differed sharply, however, in their socioeconomic background, dietary intake, and history or incidence of nutritional deficiency diseases.

The Wetzel Grid findings over a two-year period revealed that there was no significant difference in the channel course of the two groups of children. This was contrary to expectations. By contrast, the two groups, as groups, showed a distinct difference in their respective schedules of development, the children in the test group being on slower schedules of development than those in the control group. In the two-year period, the children in the test group underwent further retardation in their schedules of development. The extension of the retardation over the two-year period occurred despite their slower schedules of development. Slower schedules result in lower levels of development; this in turn suggests, concomitantly, reduced caloric needs. However, the retardation can be explained only by some inadequacy, since the children lost further ground. The dietary inadequacies seem to be the most logical explanation, since the incidence of illness was not increased in the test group.

A point of distinction between the two groups was the difference in the regularity of growth. The children in the test group, despite their slower

schedules of development, had a marked tortuous mode of up-channel progress. Wetzel has proposed that simultaneous losses in channel and schedule are Grid criteria suggestive of nutritional interference with growth and development as he defines them.[8] The difference in the number of children in the test group (eighteen) and the number of children in the control group (eleven) who experienced these simultaneous changes throughout the two-year period was not as dramatic as might have been expected. The differences may be accounted for on the basis of infection and the interrelation between schedule of development and caloric needs. The comparatively lower metabolic needs of the children in the test group were dependent on their slower schedule of development rather than on their physical condition. This permitted them to maintain their physique channel positions more readily than children on fast schedules who reach higher developmental levels.

## SUMMARY AND CONCLUSIONS

1. Medical, dietary, and Wetzel Grid findings in children without symptoms of nutritional deficiency diseases on a fairly adequate diet and children with signs and symptoms suggestive of nutritive failure were compared for a two-year period. No therapy was given during the two years.

2. The analysis of physical status by the Wetzel technique paralleled the medical and dietary observations; in general, nutritive failure in children resulted in instability of up-channel progress on the Grid. The analysis of physical progress by the Grid technique raised some equivocal questions.

3. The auxodrome as a tool proved to be a good screening device in the Nutrition Clinic for separating children into groups according to their schedules of development and regularity of progress.

4. The findings in this study revealed to us the desirability of supplementing the auxodrome with other devices, such as the x-ray findings, for the measurement of rate of growth. X-rays are necessary aids in the differentiation between children who are often grouped into a single category for two reasons; i.e., they were small either because of their genetic inheritance or small because of inadequate nutrition.

We wish to acknowledge our indebtedness to Dr. Norman Glass and the late Dr. Charles Bray, Medical Department, American Cast Iron Pipe Company, Birmingham, Ala., for their cooperation in providing us with a source of control patients.

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# FURTHER OBSERVATIONS ON POST-TRACHEOTOMY, MEDIASTINAL EMPHYSEMA, AND PNEUMOTHORAX

GILBERT B. FORBES, M.D., ST. LOUIS, MO., GEORGE SALMON, M.D., HOUSTON, TEXAS, AND JOHN C. HERWEG, M.D., ST. LOUIS, MO.

THE occurrence of two fatalities from the hitherto unexpected post-tracheotomy complications of mediastinal emphysema and pneumothorax at the St. Louis Children's Hospital several years ago[1] prompted a study in regard to the incidence and etiology of these phenomena. For this purpose we have closely observed the postoperative course of all tracheotomized patients at both the St. Louis Children's and St. Louis City Hospitals during the period from December, 1941, through May, 1946. In addition, we have endeavored to correlate such factors as the patient's age, the disease for which the tracheotomy was done, the degree of dyspnea present at the time of operation, and the procedure of preoperative bronchoscopy with the incidence of these complications.

At the time our first cases were recognized, we postulated, as have others,[2-4] that mediastinal emphysema resulted from the factor of forced inspiration in the face of an inadequate airway and an open neck wound (the tracheotomy), pneumothorax, in turn, being due to rupture of the mediastinal pleura through overdistension of the mediastinum with air. Preoperative bronchoscopy was suggested as a means of obviating these complications, and, in conference with the Department of Otolaryngology, it was decided to employ this procedure more or less routinely in future patients with a view toward determining its value. In order to test the postulate that air can make its way into the mediastinum from the cervical region, certain animal experiments were designed in which an attempt was made to produce mediastinal emphysema under conditions simulating those present in the human patient whose airway is partially obstructed at the time of tracheotomy.

Our clinical and experimental observations will be described separately.

## CLINICAL OBSERVATIONS

The cases reported here consist of ninety-nine tracheotomies done on ninety-six patients at the St. Louis Children's Hospital and twenty-one tracheotomies at the St. Louis City Hospital. Three patients are included who were previously reported.[1] Of the 120 operations in this series, seventy-six were performed by thirteen different otolaryngologists who were on either the visiting or full-time staffs of the two hospitals, thirty-six by the resident house officers in otolaryngology, and eight by the general surgeons. Of the patients, 68 per cent were 2 years of age or under, 22 per cent between 2 and 5 years, and 10 per cent between 5 and 10 years. The majority of the operations were done to relieve

From the Department of Pediatrics, Washington University School of Medicine, and the St. Louis Children's and City Hospitals.

acute respiratory distress of varying degree. All patients were carefully observed throughout the postoperative period for symptoms and signs of mediastinal emphysema and pneumothorax. Frequent physical examinations of the chest were made, and whenever possible daily fluoroscopic or radiological examinations were carried out, the latter in both the posterior-anterior and lateral views.

Of the ninety-nine operated patients observed at the St. Louis Children's Hospital, it was possible to check fifty-five by means of lateral roentgenographic examination for the presence or absence of mediastinal emphysema, and at the St. Louis City Hospital nineteen of the twenty-one patients were similarly checked. Although we cannot be certain that the remaining patients did not have mediastinal emphysema of mild degree, they exhibited no symptoms or physical signs of that disorder. Sixty-two patients at Children's Hospital and twenty at the City Hospital were checked roentgenographically for the presence or absence of pneumothorax. The remaining patients did not have clinically detectable pneumothorax. Because we were unable to determine unequivocally the presence or absence of these complications in all of our patients, we have calculated our frequency data on the basis of both the total patients in the series and the number accurately checked.

Table I gives data on the incidence of mediastinal emphysema (ME)* and pneumothorax (PT)* in our series of patients. The frequency with which ME occurred is rather striking and is much greater than we had anticipated at the beginning of the study. If we consider the entire group of patients, ME was observed in 25 per cent, but if we take only those patients who were carefully checked for this complication, the incidence becomes 41 per cent. Pneumothorax occurred in 10 per cent of the patients, considering the group as a whole, and in 15 per cent of patients who were checked roentgenographically.

TABLE I. INCIDENCE OF MEDIASTINAL EMPHYSEMA AND PNEUMOTHORAX

| HOSPITAL | NO. CASES | NO. CHECKED FOR ME | NO. ME | NO. CHECKED FOR PT | NO. PT |
|---|---|---|---|---|---|
| St. Louis Children's | 99 | 55 | 21 | 62 | 7 |
| St. Louis City | 21 | 19 | 9 | 20 | 5 |
| Totals | 120 | 74 | 30 | 82 | 12 |

Since not all patients in this series were bronchoscoped prior to tracheotomy, it is of interest to note what effect an indwelling bronchoscope at the time of operation had on the incidence of postoperative ME and PT. It was postulated at the beginning of this study[1] that the provision of an adequate airway (by means of an indwelling bronchoscope, or intubation tube) during the tracheotomy procedure would minimize the likelihood of ME and PT. The natural differences of opinion among the hospital staff in regard to the advisability of this procedure provided us with a sufficient number of patients for "controls" to allow a fairly accurate evaluation of the worth of this procedure. In several of the sixty-nine patients who were not bronchoscoped preoperatively, the

*These abbreviations will be used freely throughout the remainder of the text.

TABLE II. RELATIVE FREQUENCIES OF MEDIASTINAL EMPHYSEMA AND PNEUMOTHORAX IN RELATION TO PREOPERATIVE BRONCHOSCOPY

| HOSPITAL | PREOPERATIVE BRONCHOSCOPY | NO. CASES | NO. CHECKED FOR ME | NO. ME | NO. CHECKED FOR PT | NO. PT |
|---|---|---|---|---|---|---|
| St. Louis Children's | Bronchoscoped preoperatively | 41 | 23 | 6 | 27 | 0 |
| | Not bronchoscoped | 58 | 32 | 15 | 35 | 7 |
| St. Louis City | Bronchoscoped preoperatively | 10 | 9 | 3 | 10 | 1 |
| | Not bronchoscoped | 11 | 10 | 6 | 10 | 4 |
| Totals | Bronchoscoped preoperatively | 51 | 32 | 9 | 37 | 1 |
| | Not bronchoscoped | 69 | 42 | 21 | 45 | 11 |

TABLE III. AGE DISTRIBUTION OF PATIENTS

| AGE (YR.) | NO. CASES | NO. BRONCHOSCOPED | NO. CHECKED FOR ME | NO. ME | NO. CHECKED FOR PT | NO. PT |
|---|---|---|---|---|---|---|
| 0–1 | 34 | 16 | 20 | 7 | 22 | 3 |
| 1–2 | 48 | 24 | 33 | 14 | 38 | 6 |
| 2–5 | 26 | 10 | 14 | 8 | 15 | 2 |
| 5–10 | 12 | 1 | 7 | 1 | 7 | 1 |
| Total | 120 | 51 | 74 | 30 | 82 | 12 |

TABLE IV. DEGREE OF RESPIRATORY DISTRESS PRESENT AT TRACHEOTOMY

| DEGREE RESPIRATORY DISTRESS | NO. CASES | NO. BRONCHOSCOPED | NO. CHECKED FOR ME | NO. ME | NO. CHECKED FOR PT | NO. PT |
|---|---|---|---|---|---|---|
| None | 6 | 0 | 2 | 1* | 2 | 0 |
| Moderate | 63 | 26 | 33 | 9 | 37 | 2 |
| Marked | 38 | 19 | 29 | 15 | 32 | 7 |
| Extreme (Emergency) | 13 | 6 | 10 | 5 | 11 | 3 |

*Marked struggling.

procedure was considered advisable but not carried out because of either undue laryngeal edema or the emergency nature of the situation. Table II illustrates the relative frequencies of ME and PT in the two groups of patients, the incidence of ME being approximately 70 per cent greater for the nonbronchoscoped than for the bronchoscoped group, and the incidence of PT some eight times greater for the former group. These differences are statistically significant.*

In Table III our patients are arranged according to age. The number of patients who were bronchoscoped preoperatively is indicated for each age group in order to exclude the possibility of the comparative frequencies being distorted by that factor. Patients in the 5- to 10-year age group had the lowest incidence of ME and PT; the small number of patients in this group, however, does not allow a valid comparison.

In Table IV the patients are grouped with respect to the degree of respiratory distress present at the time of tracheotomy. Except for the group

*For the bronchoscoped and nonbronchoscoped patients, respectively, the frequencies of the occurrence of pneumothorax and the standard error of the ratios are 1.96 per cent ± 1.93 per cent, and 15.9 per cent ± 4.4 per cent. Similarly, for the occurrence of mediastinal emphysema, they are 17.7 per cent ± 5.26 per cent, and 30.4 per cent ± 5.57 per cent for the two groups of patients.

labeled "none" the percentage of preoperative bronchoscopies was about the same in each group. The incidence of ME rises from an average of approximately 15 per cent in the patients who had little or no respiratory distress to 40 per cent in those with more marked degrees of embarrassment. Similarly, the frequency of PT becomes greater with increasing respiratory distress to the point where, in the group of "emergency" cases, it occurred in 23 per cent of the patients.

Table V lists the diseases present in our patients requiring tracheotomy. The category labeled "other upper respiratory obstruction" includes such occurrences as laryngeal or tracheal foreign body, laryngeal edema (other than that resulting from bronchoscopy), granulation tissue in the trachea, and papillomas of the larynx. In the "elective" group is one patient in whom tracheotomy was done in order to administer a general anesthetic for an intraoral operation and five in whom it was done prior to a diagnostic bronchoscopy; none had respiratory distress. Of the first five groups, which are the only ones containing enough cases for accurate comparison, the cases of laryngotracheobronchitis have the highest incidence of ME. Pneumothorax occurred with greatest frequency in groups I, IV, and V; the differences in frequency for the various groups is much less striking, however, than those for ME.

TABLE V. DISEASES PRESENT IN PATIENTS REQUIRING TRACHEOTOMY

| GROUP | DISEASE | NO. CASES | NO. BRONCHO-SCOPED | NO. CHECKED FOR ME | NO. ME | NO. CHECKED FOR PT | NO. PT |
|---|---|---|---|---|---|---|---|
| I | Laryngotracheobronchitis | 26 | 12 | 19 | 11 | 20 | 4 |
| II | Acute laryngitis, diphtheria | 28 | 14 | 20 | 6 | 22 | 2 |
| III | Postbronchoscopy | 23 | 9 | 11 | 3 | 13 | 1 |
| IV | Acute bronchiolitis | 14 | 8 | 8 | 3 | 9 | 2 |
| V | Other upper respiratory obstruction | 14 | 5 | 8 | 3 | 8 | 2 |
| VI | Intraoral obstruction | 6 | 2 | 4 | 3 | 6 | 1 |
| VII | Dyspnea from pulmonary disease | 3 | 0 | 2 | 0 | 2 | 0 |
| VIII | Elective | 6 | 1 | 2 | 1* | 2 | 0 |
| | Totals | 120 | 51 | 74 | 30 | 82 | 12 |

*Marked struggling.

Of the 120 operated patients, thirty-one suffered one or both of these complications. In compiling this total, we excluded one patient who developed ME after having had a tracheotomy tube in place for twenty-seven days and who at autopsy was found to have an extensive ulceration of the trachea. Mediastinal emphysema without accompanying PT occurred in nineteen patients; eleven had both ME and PT, and one had PT alone. Three of the patients in this series were subjected to two separate tracheotomies. One, who developed ME and left PT following a tracheotomy at 2 months of age, failed to develop either complication when the procedure was repeated under identical conditions one year later. Another patient 14 months of age, developed ME and left PT following tracheotomy, only to have ME and right PT recur

following a second tracheotomy two weeks later. The third patient suffered neither complication following two tracheotomies done two weeks apart.

In those patients who suffered embarrassment from ME and/or PT, it is possible to determine fairly accurately the elapsed time from the operation to the onset of these complications. This time varied from twenty minutes to twelve hours, with an average of 3.8 hours. The time required for ME to disappear varied from two to eleven days, with an average of 6.7 days. The "precordial crackle," a peculiar crackling sound synchronous with the heart beat, and first emphasized by Hamman[5] as an important diagnostic sign in ME, was heard in only six of our patients. This sign may have been present though not detected in some patients, due to the necessity of carrying out the examination of the chest in the supine position. Subcutaneous emphysema was noted in eleven of the patients who exhibited ME. Subcutaneous emphysema also occurred in several patients who were free from ME.

Not all of the patients with ME experienced symptoms from this condition. Of the nineteen patients with ME alone (see Table VI) twelve had no untoward symptoms clearly attributable to this complication, the condition being discovered either by roentgenographic or post-mortem examination. Two patients had moderate dyspnea requiring the administration of oxygen, and three patients had severe embarrassment, resulting in the death of two of them. In the remaining two patients it was impossible to determine what part, if any, the presence of ME played in their symptoms. One was an instance of hemolytic streptococcal laryngotracheobronchitis with membrane formation in the trachea; the patient was extremely toxemic and expired twelve hours postoperatively. The other patient had diphtheria complicated by myocarditis and extensive membrane formation throughout the greater part of the tracheobronchial tree.

TABLE VI. SYMPTOMS FROM MEDIASTINAL EMPHYSEMA

| SYMPTOMS | NO. CASES |
|---|---|
| None | 12 |
| Moderate dyspnea | 2 |
| Severe embarrassment | 3 |
| Impossible to evaluate | 2 |

Twelve patients had PT; in all but one of these ME was also present (Table VII). Pleural aspiration was carried out in three of the four patients who had moderate distress with cyanosis; relief was immediate, and in only one did the aspiration have to be repeated. The other patient in this group responded satisfactorily to oxygen therapy, the air disappearing from the pleural cavity over a period of seven days. This patient, in addition to the one who

TABLE VII. MEDIASTINAL EMPHYSEMA AND PNEUMOTHORAX

| SYMPTOMS | NO. CASES | LOCATION | NO. CASES |
|---|---|---|---|
| None | 1 | Right | 5 |
| Mild | 1 | Left | 2 |
| Moderate | 4 | Bilateral | 5* |
| Severe | 6* | | |

*One patient did not have ME.

TABLE VIII.

| PATIENT | YR. | AGE | DISEASE | ELAPSED TIME | SYMPTOMS | ME | PT | TREATMENT | DISAPPEARANCE OF AIR | OUTCOME |
|---|---|---|---|---|---|---|---|---|---|---|
| *Children's Hospital* | | | | | | | | | | |
| (1) D. A. | '41 | 14 mo. | postbronchoscopy | 20 min. | gasping respirations sudden death | 0 | bilat. | recognized at death | | D |
| (2) E. A. | '42 | 12 mo. | F.B.* trachea | 4 hr. | Cheyne-Stokes, retraction, circulatory collapse | ++++ | 0 | aspiration mediastinum failed | | D |
| (3) L. A. | '42 | 2 mo. | hemangioma soft palate | 20 min. | dyspnea, cyanosis | ++ | right | aspiration pleura twice | 3 days | R† |
| (4) T. B. | '42 | 14 mo. | L-T-B‡ | 6 hr. | dyspnea, retraction, mediastinal shift | ++ | left | aspiration pleura once | 3 days | R |
| (5) G. B. | '43 | 14 mo. | acute laryngitis | 7 hr. | dyspnea, mediastinal shift | + | left | aspiration pleura once | 8 days | R§ |
| (6) S. W. | '45 | 2 yr. | F.B.* trachea | 2 hr. | retraction, cyanosis, marked distress | + | bilat. | aspiration right pleura twice, left once | 7 days | R |
| (7) R. B. | '46 | 23 mo. | L-T-B | 45 min. | cyanosis, dyspnea | ++++ | 0 | repeated aspiration mediastinum | 9 days | R |
| *City Hospital* | | | | | | | | | | |
| (8) L. M. | '42 | 2 mo. | bronchiolitis | 2 hr. | repeated attacks cyanosis, dyspnea, death in 8 hr. | + | right | undiagnosed | | D |
| (9) W. W. | '42 | 4 mo. | bronchiolitis | 6 hr. | repeated attacks dyspnea, retraction, sudden death in 6 hr. | +++ | right | undiagnosed | | D |
| (10) G. L. | '43 | 18 mo. | acute laryngitis | 12 hr. | faulty tube, then hypopnea, cyanosis, gasps | ++++ | 0 | undiagnosed | | D‖ |
| (11) L. E. | '44 | 7 yr. | L-T-B | 30 min. | shallow rapid respirations | + | bilat. | oxygen | 7 days | R |
| (12) R. I. | '44 | 3½ yr. | acute laryngitis | 8 hr. | sudden death | ++++ | bilat. | none | | D |
| (13) P. B. | '45 | 14 mo. | L-T-B | 2 hr. | retraction, cyanosis, dyspnea, marked distress | +++ | bilat. | continuous siphon suction both pleurae, unsuccessful | | D |

*Foreign body.
†Tracheotomy one year later without complication.
‡Laryngotracheobronchitis.
§Recurrence of M.E. and P.T. following second tracheotomy two weeks later.
‖Retroperitoneal emphysema also.

exhibited only mild symptoms (increased respiratory rate), would not have been diagnosed had not routine roentgenographic examination been done.

Since ME and PT can occur in nontracheotomized patients suffering respiratory embarrassment and from causes unknown in otherwise healthy individuals, the question may be raised as to whether these complications could have been present prior to tracheotomy in some of the patients in our series. In sixteen of our thirty-one cases of ME and/or PT, preoperative fluoroscopy was done,

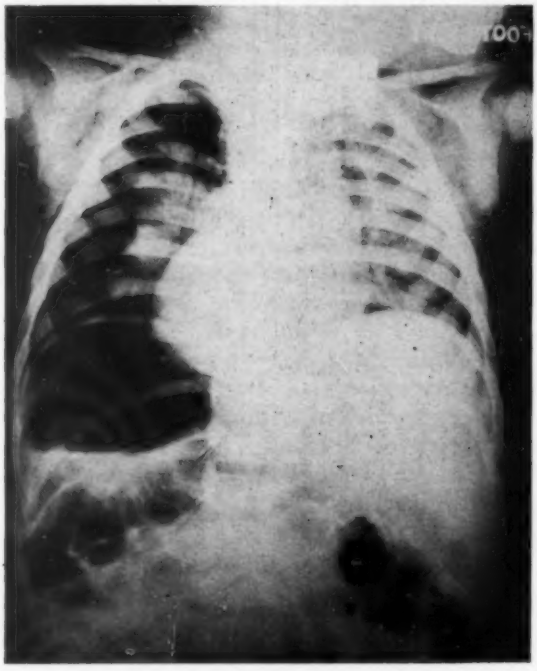


Fig. 1.—Case 5. Posterior-anterior roentgenogram showing mediastinal emphysema, left pneumothorax, and subcutaneous emphysema. Note that the film was reversed during the printing process.

and it is definitely known that these conditions were not present prior to tracheotomy. Of the remaining fifteen patients, seven had symptoms from ME and/or PT during their postoperative course following the initial relief due to the tracheotomy. There was one patient who developed ME following tracheotomy and yet had no respiratory distress whatsoever prior to the operation (Table IV).

The patients who exhibited notable symptoms from ME and PT are presented in more detail in Table VIII. The two hospitals are separated in the table, the patients at each being listed in chronological order. It should be noted that three patients suffered marked embarrassment from ME alone; two

of them died, and the other most certainly would have, had mediastinal aspiration not been done. In order to illustrate the symptomatology and handling of these patients, several of the more interesting ones will be presented in detail. Cases 1, 2, and 3 have been reported previously.[1]

Fig. 2.—Lateral roentgenogram showing extensive mediastinal emphysema (Case 10).

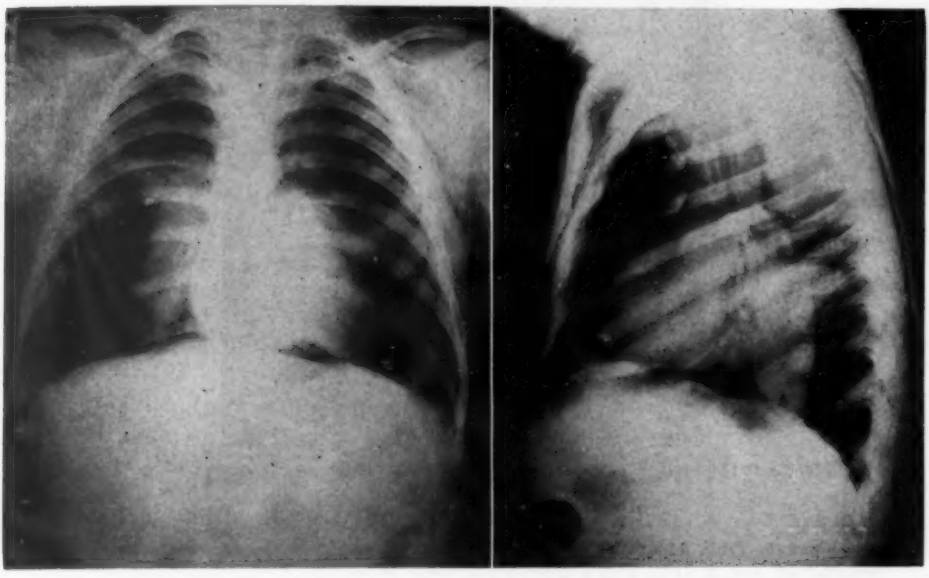
CASE REPORTS

CASE 5.—This 14-month-old white male was admitted to the St. Louis Children's Hospital on Aug. 3, 1943, with a diagnosis of acute laryngitis. Tracheotomy afforded considerable relief. On the following day the infant exhibited moderate dyspnea without cyanosis or other symptoms. Physical signs of left PT were first noted on this day, and the chest roentgenogram (Fig. 1) revealed the findings at that time. From the left pleural cavity, 28 c.c. of air (under pressure) were aspirated with resultant respiratory relief and shifting of the heart back to its normal position. The remaining air disappeared completely over a period of eight days. Interestingly enough, two weeks later a second tracheotomy was performed, followed in several hours by ME and left PT. Pleural aspiration was not required on this occasion as no symptoms were noted; the air disappeared spontaneously over a period of eleven days.

This case is illustrative of those with mild symptoms for whom a pleural aspiration affords substantial and permanent relief. Even though small amounts

of air were removed, the intrapleural pressure was sufficiently reduced to ameliorate symptoms.

CASE 10.—This 18-month-old white female was admitted to the St. Louis City Hospital Jan. 12, 1943, suffering from laryngotracheobronchitis. Tracheotomy produced considerable relief. The patient did fairly well until twelve hours postoperatively when the tracheotomy tube became slightly misplaced and filled with mucus. She suffered extreme distress at this time and was revived with considerable difficulty. Bronchoscopic examination revealed the tracheobronchial tree to be free of obstruction. Despite repeated tracheal suction and oxygen therapy, she continued to have repeated episodes of dyspnea and cyanosis. Respirations gradually became more shallow; cyanosis increased, and she expired about an hour after the original episode. Fig. 2 is a roentgenogram taken shortly after death, showing massive ME. Subcutaneous emphysema and PT were not present. Autopsy disclosed an extensive ME which was estimated to fill approximately one-half of the thoracic cage. In addition, there was considerable retroperitoneal emphysema extending down to the common iliac vessels.



*A.* *B.*

Fig. 3.—*A*, posterior-anterior, and *B*, lateral roentgenograms of the chest (Case 12).

CASE 12.—This 3-year-old white male was admitted to the St. Louis City Hospital on Oct. 24, 1944, with acute laryngitis. There was marked retraction on inspiration. Tracheotomy was done with difficulty because of much struggling. He was relieved by this procedure. Thirty minutes postoperatively fluoroscopy revealed questionable ME. During the next few hours he had two or three attacks of moderate dyspnea and cyanosis, relieved in each case by suctioning mucus from the tracheotomy tube. Eight hours postoperatively he suddenly expired. Examination of the chest revealed signs of bilateral pneumothorax, and roentgenograms were made (Fig. 3*A* and *B*) which show extensive ME and bilateral PT. Subcutaneous emphysema was also present. These findings were confirmed at autopsy, at which time pleural aspirations demonstrated the pneumothorax to be of the tension variety.

These two cases are illustrative of what happens when ME and PT are neither diagnosed nor suspected. In Case 10 it is probable that ME was initiated, or at least intensified greatly, by the malposition and plugging of the tracheotomy tube. Had fluoroscopic examination of the chest been done at that time, ME would have been diagnosed and the symptoms attributed to that rather than to tracheal mucus or some other pulmonary difficulty. In Case 12 it is obvious that PT, of the extent shown in Fig. 4, could not have occurred during the few moments before death, and that had careful examinations of the chest been done during the early postoperative course, PT would have been diagnosed sooner.

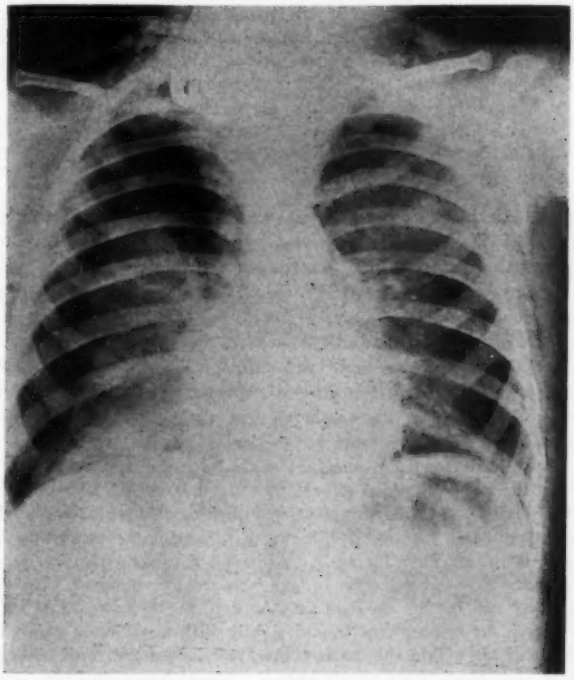


Fig. 4.—Posterior-anterior roentgenogram of the chest taken the day following bilateral pleural aspiration, showing persistence of right pneumothorax (Case 6).

CASE 13.—This 14-month-old white female was admitted to the St. Louis City Hospital in 1945 with laryngotracheobronchitis; respiratory retraction was pronounced. Tracheotomy afforded considerable relief although dyspnea continued due to the bronchitis. Two hours postoperatively dyspnea became worse, retractions increased, and cyanosis and subcutaneous emphysema was noted. Signs of ME and bilateral PT developed, and their presence was confirmed by fluoroscopy. Bilateral pleural aspiration yielded considerable amounts of air under positive pressure. Relief of symptoms was only temporary, for within just a few minutes respiratory distress was again evident. During the next five hours, until the patient's death, constant attention, oxygen and stimulants were necessary. Repeated pleural aspirations were done, and finally, because the chest seemed to fill rapidly

with air after each tap, closed siphon drainage of each pleural cavity, using an 18-gauge needle, was instituted. Air bubbled out continuously through each needle, but the patient rapidly lost ground and expired seven hours postoperatively. Autopsy confirmed the presence of massive ME and bilateral PT.

CASE 6.—This 2-year-old white male was admitted to the St. Louis Children's Hospital on Nov. 3, 1944, with a diagnosis of tracheal foreign body. He had had episodes of coughing and fever during the previous three months. Physical and roentgenographic examination of the chest revealed evidence of chronic bronchitis; ME was not present. Bronchoscopy was unsuccessful in removing the foreign body, which lodged in the larynx, necessitating an emergency tracheotomy. Repeated low bronchoscopies failed to locate the foreign body. Two hours following tracheotomy the child's condition became much worse, retractions were marked, and he was moderately cyanotic. Fluoroscopy at this time revealed ME and bilateral PT. Pleural aspiration yielded 270 c.c. of air from the right side and 170 c.c. from the left under pressure. Immediate relief resulted from this procedure. The following day subcutaneous emphysema developed, and the child seemed very restless, exhibiting an increased respiratory rate but no retraction. A chest roentgenogram at that time (Fig. 4) showed right PT. Five hundred cubic centimeters of air were withdrawn with improvement. Bronchoscopy was done later that day and the foreign body recovered from the right main bronchus. The patient's further course was uneventful, air disappearing from the mediastinum within seven days.

CASE 7.—A 23-month-old white male was admitted to the St. Louis Children's Hospital on Feb. 10, 1946, with the diagnosis of laryngotracheobronchitis and salicylate intoxication. Tracheotomy afforded relief from the respiratory obstruction, yet his general condition remained poor, and hyperpnea continued because of the salicylism. A small amount of ME was noted thirty minutes after the operation; within an additional fifteen minutes ME had become very pronounced, and subcutaneous emphysema developed. The pulse was very rapid and thready, the respirations rapid and shallow; intense cyanosis developed, and the baby appeared to be in extremis. The precordium was resonant to percussion, and a faint precordial crackle was audible. Pneumothorax was not present, nor was it noted subsequently. Stimulants, oxygen, and repeated suctioning of the tracheotomy tube failed to produce improvement. His condition remained critical for the next three and one-half hours, during which time the pulse remained above 120 though it varied in volume; blood pressure readings were 130/80 and 115/75 on two occasions. During this period repeated aspirations of the mediastinum were done, a total of 400 c.c. of air being withdrawn from the left side (of the sternum) and 10 c.c. from the right. Although the first aspiration produced considerable relief, subsequent ones did not seem to help him greatly; however, they were done whenever the amount of air in the mediastinum seemed to be increasing as determined by fluoroscopy. Packing the tracheotomy wound tightly with merthiolate gauze did not prevent the reaccumulation of air within the mediastinum, and it was not until a larger tracheotomy tube was inserted that the ingress of air ceased. Air exchange in both lungs seemed good at all times except for one occasion when the breath sounds became depressed at the left lower lung field; they quickly returned to normal following the removal of some air from the mediastinum. During the next twelve hours, three additional mediastinal aspirations were done with recovery of a total of 25 c.c. of air. He continued to exhibit periods of moderate respiratory distress during the next three days; mediastinal and subcutaneous emphysema gradually diminished, however, and his subsequent hospital course was uneventful.

These three cases illustrate treatment. Our experiences with Case 7 suggest that the treatment of Case 13 might have been successful had aspiration of the mediastinum been done. In the latter case it is evident that the closed intercostal siphon drainage system was inadequate, and it might have been better

if a larger tube had been used, so that air could have escaped from the pleural cavities as rapidly as it entered. Case 7 illustrates an unusual set of circumstances in that the salicylism resulted in continuance of respiratory difficulty after the tracheotomy had been completed. Although the tracheotomy tube was patent, it apparently did not allow sufficient respiratory exchange to satisfy the demands produced by the salicylate poisoning, and air continued to accumulate in the mediastinum until a larger tube was inserted. The life-saving effects of mediastinal aspiration are well illustrated here. These three cases leave no doubt that ME and/or PT can produce alarming symptoms.

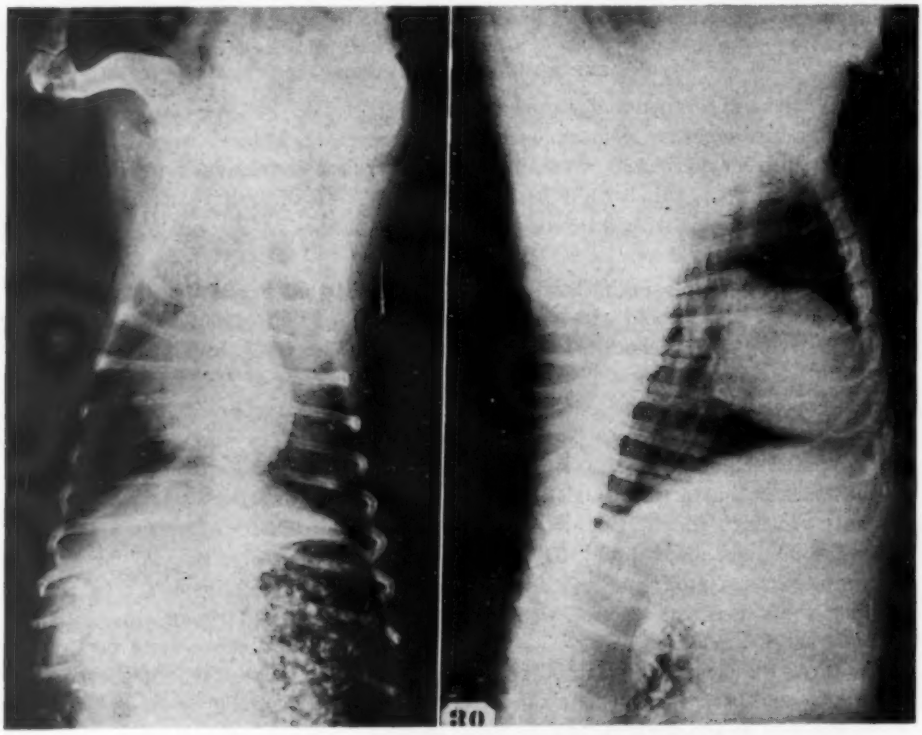
## EXPERIMENTAL OBSERVATIONS

Various experiments, designed to test the postulate that air can be drawn into the mediastinum from an open tracheotomy wound, were carried out on dogs. In all experiments a routine midline cervical incision was made after infiltration of the skin with 1 per cent procaine. In several it was necessary to supplement this with intravenous nembutal anesthesia. A total of thirteen experiments were done.

The first four experiments were done on 3-week-old puppies, weighing approximately 1 kg. each, with the idea (at the suggestion of Dr. A. J. Cone) of observing the phenomenon of tracheal spasm described by Cole[6] and of seeing whether this might play a role in the pathogenesis of ME. The trachea was exposed but not incised in these experiments. After preliminary roentgenograms were taken, the trachea and peritracheal tissues were mechanically stimulated with a hemostat. In one of the four dogs tracheal spasm was noted, the trachea contracting to about one-half normal size. Dyspnea ensued and continued until the animal was sacrificed three hours later. At autopsy there was a minimal amount of ME and no PT. Unfortunately, the animal had aspirated some milk, and this may have accounted for the dyspnea. In the other three dogs, the tracheal wound was filled with a dye (trypan blue), and the trachea was completely occluded with a hemostat for thirty-second periods on three occasions with resultant severe dyspnea and retraction. Roentgenographic and post-mortem examination failed to reveal ME or PT or the presence of the dye in the mediastinum of these animals.

The remaining experiments were carried out on adult dogs weighing 7 to 8 kg. Tracheotomy was done and a tracheal cannula inserted and secured firmly in place with a single tie about the trachea. This latter procedure, though objectionable because it obviously disturbs the peritracheal tissues more than is usual in human tracheotomy, was necessary in order to produce sufficient dyspnea upon occlusion of the cannula. The tracheotomy wound was then filled with a liquid radiopaque material or a dye and the tracheal cannula occluded to the point of profound dyspnea. Roentgenograms were taken at intervals, and after a suitable length of time the dog was sacrificed. At post-mortem examination the chest was opened under water.

*Experiment V.* Local anesthesia only: 30 c.c. of a 0.5 per cent solution of trypan blue placed in tracheotomy wound. Moderate dyspnea maintained for one hour. Roentgenograms (Fig. 5, *A* and *B*) show ME and left PT. At autopsy, air was seen to fill the superior mediastinum, and there were bubbles of air about the great vessels and along the aorta as far down as the diaphragm (Fig. 5, *C*). Trypan blue extended into mediastinum down to tracheal bifurcation. Injection of mediastinum with methylene blue solution revealed a small rupture in the left superior-posterior mediastinum. The lungs were grossly normal; no tear in visceral pleura could be detected by injection of tracheobronchial tree with methylene blue.



*A.* *B.*

Fig. 5*A*, *B*, *C*.—Experiment V. *A*, anterior-posterior and *B*, lateral roentgenograms, showing mediastinal emphysema and left pneumothorax.

*Experiment VI.* Conditions of Experiment V were repeated. Dyspnea was maintained for one hour. A moderate amount of air and trypan blue was found in the superior mediastinum at autopsy. PT was not present.

*Experiment VII.* 30 c.c. of lipiodol were placed in the tracheotomy wound. Moderate dyspnea was maintained for thirty minutes, during which time the lipiodol could be seen to disappear gradually. A roentgenogram taken at this time is illustrated in Fig. 6. Dyspnea was maintained for an additional two and one-half hours, whereupon lipiodol and air continued to collect in the mediastinum (Fig. 7, *A* and *B*). Autopsy confirmed the presence of

massive ME extending to diaphragm. There was no PT. The right lung pleura contained numerous petechial hemorrhages.

*Experiment VIII.* Conditions of Experiment VII were repeated, using thorotrast as a contrast medium. A small amount of thorotrast entered the superior mediastinum, but there was no evidence of ME and PT either roentgenographically or at postmortem examination.

*Experiment IX.* Conditions of Experiment VII were repeated, using a 1 per cent solution of trypan blue to fill the tracheotomy wound. At autopsy the trypan blue was seen to have traveled almost to tracheal bifurcation, but there was no evidence of ME or PT.

*Experiment X.* Conditions of Experiment VII were repeated. There was moderate dyspnea for one hour and no evidence of ME or PT.

Fig. 5, *C.*—Postmortem photograph showing emphysema of the superior mediastinum and occasional streaks of trypan blue in that region. The right rib cage has been cut away.

*Experiment XI.* Conditions of Experiment VII were repeated. There was severe dyspnea for two hours and no evidence of ME or PT.

*Experiment XII.* Conditions of Experiment VII were repeated with addition of intravenous nembutal anesthesia. The animal was so depressed by the anesthetic that only mild dyspnea could be produced. Dyspnea maintained for two hours. Roentgenographic and postmortem examinations revealed a minimal amount of ME but no PT. The lungs were grossly normal.

*Experiment XIII.* Following intravenous nembutal anesthesia, moderate dyspnea was produced by inserting a holed cork in the larynx. Tracheotomy was then done in the usual manner. Roentgenograms taken immediately following tracheotomy revealed no evidence of ME or PT. A tracheal cannula was then inserted, the wound filled with a mixture of lipiodol and 1 per cent trypan blue solution, and the cannula occluded so as to produce moderate dyspnea. Roentgenograms were taken after fifteen minutes (Fig. 8, *A* and *B*). The dog was sacrificed after ninety minutes. Autopsy revealed considerable ME but no PT. Intimately mixed with the air in the superior mediastinum are to be seen both the trypan blue and lipiodol (Fig. 9, *A* and *B*).

## DISCUSSION

A number of instances of mediastinal emphysema and pneumothorax, occurring both in association with tracheotomy and spontaneously, have now been reported, and an analysis of these phenomena has been presented in two excellent reviews.[3, 7] It is evident from a study of our data that both ME and PT are seen with considerable frequency in tracheotomized patients, the

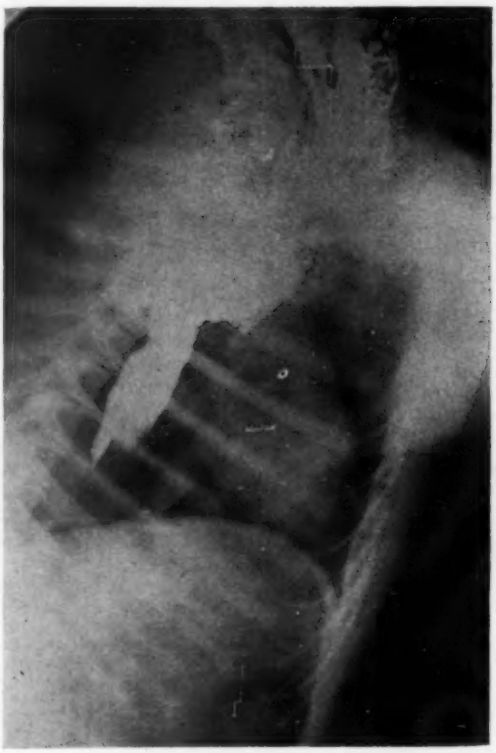
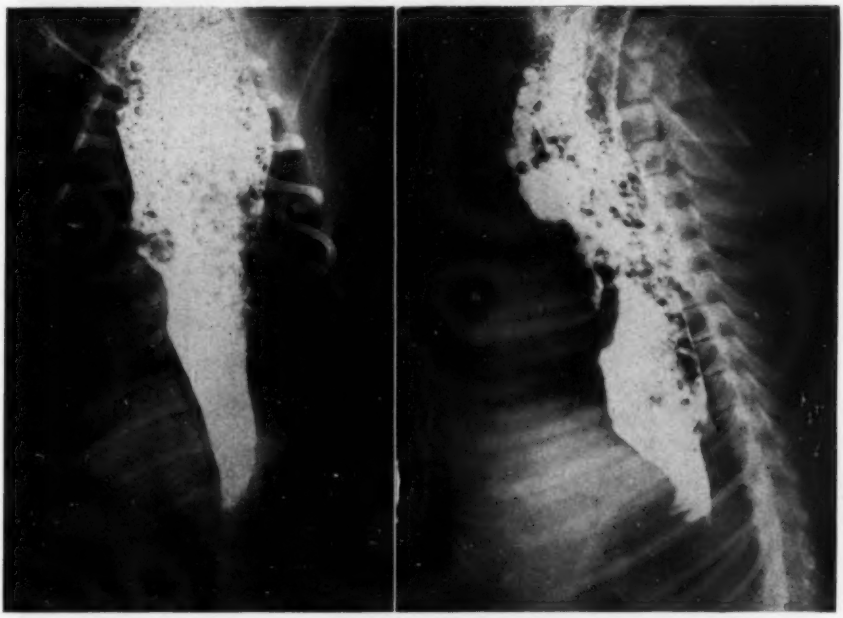


Fig. 6.—Experiment VII. Lateral roentgenogram taken after thirty minutes of dyspnea, showing lipiodol in superior and posterior mediastinum.

percentages being 25 and 10 respectively in our series.* The contention that experience of our clinic in this regard might be unique is disproved by two other reported series of tracheotomized patients. Neffson,[3] reviewing 126 cases of nondiphtheritic obstruction of the larynx, trachea, and bronchi from the Willard Parker Hospital, found seventeen instances of PT and thirty of ME, which compares favorably with our experience. He ascribes death due to tension PT in five cases; in two of these ME was not present. In one case PT (without ME)

*When only those patients who were accurately checked are considered, ME occurred in 41 per cent and PT in 15 per cent of our patients.

occurred before the trachea was incised. Goldberg and associates[4] found twelve instances of ME and eight of PT in a series of eighteen tracheotomized patients. It is entirely possible that many instances of both ME and PT were missed or their importance minimized in former days when diphtheria was a common indication for tracheotomy.

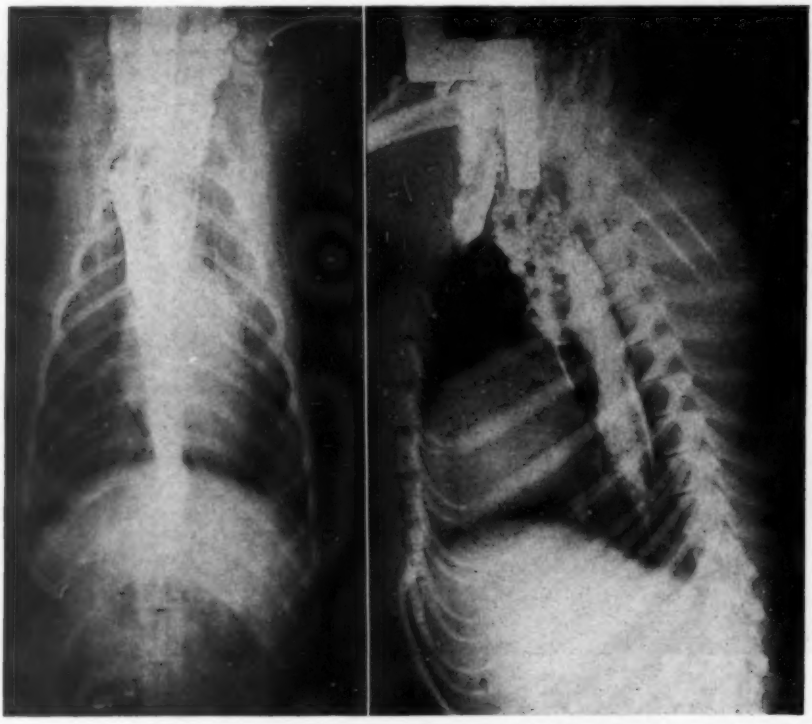


*A.* *B.*

Fig. 7.—Experiment VII. *A*, anterior-posterior and *B*, lateral roentgenograms taken after an additional two and one-half hours of dyspnea, illustrating the extensive collection of air and lipiodol in that mediastinum.

The fact that ME and PT can occur in nontracheotomized patients raises the question of whether the tracheotomy might be merely coincidental and not a cause of these conditions. Might not the ME have been present prior to the tracheotomy? The Macklins[7] take this view and state that although air may in some cases enter the mediastinum via the cervical fascial planes, the "intrinsic" route (minute alveolar ruptures leading to pulmonary interstitial emphysema in turn leading to ME) is the most common. Although we do not have accurate preoperative data for all our patients with ME and PT, we believe the occurrence of these complications to be so intimately connected with the procedure of tracheotomy as to leave little doubt of a cause and effect relationship. In certain of our patients we are positive that ME and PT were absent prior to tracheotomy. Further, although we do not doubt that ME can occur in the way proposed by the Macklins, we have not witnessed a single

case of ME in a nontracheotomized patient (with the exception of the newborn) with severe respiratory embarrassment due to asthma, tracheobronchial foreign body, mediastinal tumor, pneumonia, bronchiolitis, and the like during the past five years.

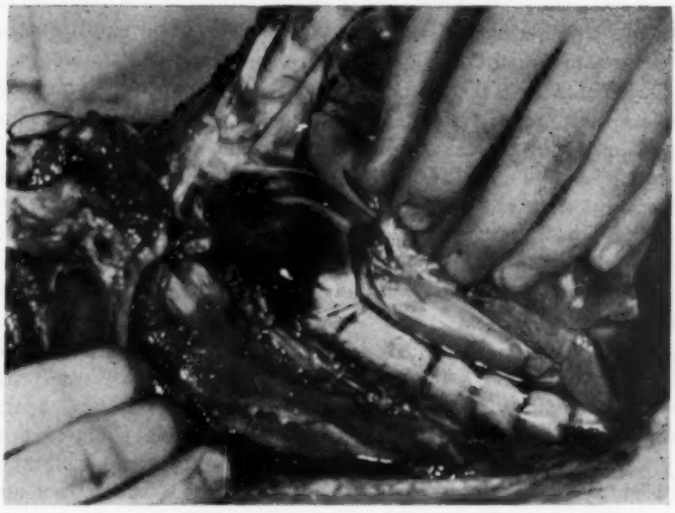


*A.* *B.*

Fig. 8.—Experiment XIII. *A*, anterior-posterior and *B*, lateral roentgenograms showing air and lipiodol in the mediastinum.

From the available evidence there are only two ways in which ME can be produced: the "extrinsic" route, wherein air is drawn into the mediastinum via the cervical fascial plans and the "intrinsic" route in which air makes its way from minute alveolar ruptures along the pulmonary vascular sheaths to the mediastinum. The Macklins favor the latter explanation and state that it may occur whenever there is an atelectasis with compensatory hyperinflation of the lung, in instances of general hyperinflation of the lung with or without increased alveolar pressure, and whenever there is a decreased blood supply to the pulmonary vessels.

It has been amply demonstrated that ME and PT can be produced experimentally in a number of ways. The Macklins have produced ME and PT by introducing air under pressure into the tracheobronchial tree in nontracheotomized cats, and from their experiments developed their theory of the



*A.*

*B.*

**Fig. 9.—Experiment XIII. Postmortem photographs. *A*, air and trypan blue in superior mediastinum. *B*, with the right shoulder and the first layer of neck muscles removed, the collection of air and trypan blue is seen to be continuous from the neck region into the superior mediastinum.**

causation of pulmonary interstitial emphysema and its attendant complications. Ballon and Francis[8] produced PT by overdistending the mediastinum with air. Goldberg and his associates[4] demonstrated that mediastinal rupture occurred in the human cadaver when the mediastinal pressure was raised to 40 mm. of mercury. They noted that the mediastinal tear occurred most frequently in the region of the left inferolateral border of the thymus. Griffin[9] produced ME and PT by subjecting the chests of tracheotomized cats to decreased atmospheric pressure. DeCosta[10] found that bullous ME could be produced in dogs on some occasions by inflating the lung with pressures of 18 to 20 mm. of mercury; in other experiments he failed to do so with pressures as high as 30 mm. High inflation pressures (60 to 90 mm.) produced massive air embolism and sudden death. We have not observed air embolism in our patients or experimental animals.

Our experiments have conclusively shown that, in the dog, air can be drawn into the mediastinum from the cervical region and that PT can result from rupture of the mediastinal pleura under conditions which prevail both during and after human tracheotomy; that is, upper respiratory obstruction. The pathway taken by the air has been traced accurately by the use of trypan blue and lipiodol, and there is no doubt that it can come from above under the conditions of our experiments. In certain animals, ME resulted from only a few minutes' dyspnea; in others, it did not occur after several hours of continued respiratory distress. Pneumothorax occurred in only one animal. In this regard, the experimental animals behave much as do human patients, for ME and PT do not occur in every tracheotomized patient. A factor which remains to be determined is the critical mediastinal pressure necessary to produce rupture of the mediastinal pleura (Goldberg's experiments suggest that it must be above 20 mm. of mercury).

Our clinical data clearly show the advisability of preoperative bronchoscopy (or any other method of assuring an adequate airway during tracheotomy, such as intubation). Neffson[3] found that the incidence of PT was reduced from 18 per cent to 12 per cent through the use of preoperative intubation. Our own figures for the bronchoscoped and nonbronchoscoped patients, respectively, are 28 per cent and 50 per cent for ME and 3 per cent and 24 per cent for PT. The data also show a good correlation between the degree of respiratory distress and incidence of both ME and PT. It is interesting and significant that laryngotracheobronchitis, which was the disease present in 22 per cent of the patients in our series, accounted for 37 per cent of our cases of ME and 30 per cent of those of PT. There are two reasons for this. In the first place, the disease, involves the entire respiratory tract, so that even though laryngeal obstruction is relieved by tracheotomy, some dyspnea may continue to occur; the thick secretions present in the tracheobronchial tree make it more difficult to keep the tracheal airway free. Secondly, the extreme dyspnea and widespread pulmonary pathological changes operate to favor the production of pulmonary interstitial emphysema according to the theory of Macklin and Macklin.[7] The differences in the incidence of ME and PT in cases of laryngotracheobronchitis

and those of pure laryngeal obstruction (where obstruction is completely relieved by tracheotomy) is evident from a study of Table V.

The age factor was also considered in our study. Not only do young children tend to suffer more acute degrees of respiratory obstruction than do older individuals, but they also present more of a technical problem to the surgeon. The operative field is smaller, thus tempting the surgeon to use a lower incision than usual (in at least one of our patients it was felt that the upper mediastinum was entered during the operation), and the patient is less cooperative and more inclined to struggle. The fact that infants have a very short neck may also be of importance, for it has been observed that ME following extensive neck dissections is somewhat more common in short-necked than in long-necked individuals.[11] Neffson states that PT occurred in his cases most frequently in the 1- to 2-year age group. Our data show no clear-cut preponderance of cases in any of the younger age groups although they do suggest that ME and PT are less likely to occur in older children.

The practical implications of our clinical observations and experimental work are clear. Efforts should be made to assure an adequate airway at all times in tracheotomized patients. The time of operation is a critical one, for it is then that the cervical fasciae are first opened and provision made thereby for the entry of air into the mediastinum. Tracheotomy should be performed as early as possible, once its need becomes apparent. If the operation can be carried out before the patient reaches the stage of extreme dyspnea, the procedure can be done less hurriedly and, consequently, more skillfully, and the likelihood of ME and PT is appreciably reduced (see Table IV). Further, in those patients exhibiting dyspnea, the assurance of an adequate airway (by means of an indwelling bronchoscope or intubation tube) at the time of tracheotomy does much to obviate the occurrence of these complications. A technical point of importance is the avoidance of lifting the pretracheal fascia away from the trachea preparatory to incising the trachea. This maneuver, through opening up another cervical fascial plane, would seem to predispose to the production of ME. The neck incision should be made as high as possible without endangering the first tracheal cartilage. If the incision is high, then retraction on inspiration may compress the lower neck tissues sufficiently to help prevent the dissection of air down the cervical fascia. In the low operation the risk of actually entering the superior mediastinum during the incision is a real one. The suggestion has been made of tightly packing off the tracheotomy wound in order to prevent the occurrence of ME. We agree with Neffson that this procedure is not indicated. It was attempted once (see Case 7) without success. It is obviously impossible to make the wound air tight by packing it with gauze, and the likelihood of infection of the wound is increased by such a procedure.

Vigilance should not be relaxed during the postoperative period, for if anything happens to obstruct the airway, ME can occur at any time. Special nursing care should be available, particularly in young children, to insure a patent, properly placed tube at all times. Case 10 illustrates the consequences of a misplaced mucus-filled tracheotomy tube. Concurrently, efforts should be made,

in those patients who have lower respiratory distress as well as laryngeal obstruction (such as laryngotracheobronchitis), to promote a good exchange of air at the alveolar level. The use of an atmosphere of humidified air, the frequent inhalation of carbon-dioxide-oxygen mixtures, the intratracheal instillation of a specially prepared physiological solution* together with a vasoconstrictor such as neosynephrin, and the administration of the sulfonamide drugs and penicillin are all useful therapeutic adjuncts.

The symptomatology and proper handling of ME and PT should be thoroughly understood; what is most important, however, is that *these complications should be considered in the differential diagnosis whenever untoward symptoms develop in a tracheotomized patient.* Our statements to the effect that of the instances of ME and PT observed in our series only a certain number resulted in embarrassment to the patient should not detract from the seriousness of these complications.

"Air in the mediastinum and interstitial tissues of the lung may be occult, unrecognized by means of visible manifestations. It accompanies a wide variety of clinical conditions and respiratory diseases. Especially when it is occult, it may be malignant; when it is malignant, it can be fatal. When its presence is diagnosed, it may be withdrawn, and thus the patient's life may be saved."

Had the possibility of the occurrence of ME and PT in some of our fatal cases been kept in mind, proper treatment might have produced a different outcome.

Physical examination of the chest should be done carefully and frequently in all postoperative cases, and a fluoroscopic examination or roentgenogram made whenever possible within the first six hours. Although ME may be diagnosed from an anterior-posterior roentgenogram (it is often erroneously considered to be "pneumopericardium"), a lateral view is much more satisfactory and informative. In addition to the roentgenographic findings, the only pathognomonic sign of ME is the peculiar "precordial crackle" referred to earlier in this paper. The other symptoms and signs are not diagnostic but may be of help in alerting the observer. When mediastinal emphysema becomes sufficiently pronounced, circulatory as well as pulmonary embarrassment ensues. The venous return to the heart is impeded, so that the pulse becomes rapid and weak; the blood pressure falls, and the neck veins are distended. The patient becomes dyspneic, cyanotic, quite apprehensive, and, if the process continues, exhausted to the point of coma in his fight for air. In adults, pain of a character suggestive of coronary disease may be present, and electrocardiographic changes have been described (slight elevations of ST segments, inverted T waves).[13] The patient may even experience temporary relief when the mediastinum ruptures to produce PT. The symptoms and signs of PT are too well known to invite discussion here.

The treatment of post-tracheotomy ME and PT involves two principles: the withdrawal of the air, and the prevention of its further extension or reaccumulation. In those instances in which the process is limited in extent and

*A physiological fluid of the following composition: $NaCl$, 0.73 per cent; $KCl$, 0.04 per cent; $CaCl_2$, 0.01 per cent; $MgCl_2$, 0.02 per cent; and $NaHCO_3$, 0.2 per cent prepared as described by Hartmann.[13]

is not causing undue symptoms, application of the latter principle is all that is necessary. If liberal amounts of oxygen are provided and the airway made to function properly, the patient will usually suffer no further harm, the air being resorbed from the mediastinum or pleura in a few days. However, if air continues to accumulate within the mediastinum and/or pleural cavity to the detriment of the patient, then in addition to the above measures it must be removed. A single pleural aspiration, using an oiled syringe and a small gauge needle is often all that is needed to reduce the tension sufficiently within the pleural space and provide relief from PT (see Table VIII). If air continues to enter the pleural space, then a closed system of intercostal siphon drainage must be provided. The intercostal tube should be large enough to drain the air from the pleural space as rapidly as it enters via the mediastinum, or else the tension will not be relieved (see Case 13).

The treatment of ME is somewhat more difficult. Simple aspiration was successful in one of our patients (Case 7) and failed in another (Case 2). The technique consists of entering the second or third intercostal space just lateral to the sternum and directing the needle, attached to a sterile oiled syringe, toward the midline of the thorax. The danger of entering the heart or one of the great vessels is lessened by virtue of the fact that these structures are pushed posteriorly by the collection of air in the mediastinum. Because the air is often present as numerous varied-sized bubbles, it is necessary to explore cautiously the anterior mediastinum in order to remove any quantity of air. Cervical mediastinotomy has also been successful in the treatment of mediastinal emphysema.[14, 15]

## CONCLUSIONS

In a study of 120 consecutive post-tracheotomy cases at the St. Louis Children's and City Hospitals during the past four and one-half years, the incidence of mediastinal emphysema and pneumothorax was found to be appreciable, mediastinal emphysema being diagnosed in 25 per cent and pneumothorax in 10 per cent of the cases.

Mediastinal emphysema and pneumothorax were produced in tracheotomized dogs under experimental conditions by obstructing the tracheal cannula to the point of dyspnea. Using lipiodol and trypan blue as tracers, the course of the air followed from the tracheotomy wound into the mediastinum was outlined. In one animal, actual rupture of the mediastinal pleura with resultant pneumothorax was observed.

The value of providing an adequate airway at the time of tracheotomy, by means of an indwelling bronchoscope, in lowering the incidence of mediastinal emphysema and pneumothorax was amply demonstrated in our clinical material.

The prevention of mediastinal emphysema and pneumothorax, the diagnosis of these conditions, and their proper handling are discussed. Their potential seriousness is emphasized.

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# A STUDY OF THE ABSORPTION AND EXCRETION OF ORAL PENICILLIN IN CHILDREN

MILTON MARKOWITZ, M.D., AND ANN G. KUTTNER, M.D.
IRVINGTON-ON-HUDSON, N. Y.

AN INCREASING number of reports have been published recently which indicate that with adequate dosage, effective blood levels may be obtained following oral administration of penicillin. Early investigators, Florey and associates,[1-3] Abraham and associates,[2] and Rammelkamp and Keefer,[3] found that only small amounts of penicillin were detected in the blood and urine following oral administration, indicating that although absorption did occur, it was variable and of a small degree. Since it was known that penicillin was unstable at a low pH, it was thought that gastric acidity might account for the low blood levels.

The majority of the recent studies[4-12] have been concerned with attempts to protect penicillin against the acidity of the stomach by the use of various combinations of antacids and protective coatings. Although it is still not clear which of these vehicles is the most efficacious, most authors state that following the simultaneous administration of buffers, higher blood levels were obtained than when an equivalent amount of penicillin was given in aqueous solution. The findings of McDermott and his co-workers,[13] however, indicated that gastric acidity was probably not a major factor in the destruction of orally administered penicillin. In studying patients with achlorhydria, they found that the degree of absorption, as measured by the urinary excretion of penicillin, was no greater than that observed in subjects with normal acid secretion. In an earlier study,[14] the same authors found that following various methods of oral administration in the same fasting subject, there was no difference in the concentration attained in the blood, regardless of whether penicillin was administered with a protective agent or in aqueous solution. Similarly, Charney and associates[7] noted that when oral penicillin was given on a fasting stomach the amount of urinary excretion was increased only slightly by the simultaneous administration of a buffer substance. Finland and his co-workers[15] also reported that the blood concentrations obtained following the ingestion of penicillin in aqueous solution were as high as with any special preparation, with the possible exception of aluminum hydroxide gel, and that the relationship to meals rather than the addition of buffer was an important factor in determining the amount of absorption. Although there is still a divergence of opinion in regard to the importance of the destructive action of gastric acidity on penicillin, it is generally agreed that the presence of food in the stomach influences absorption markedly.

From Irvington House, Irvington-on-Hudson.
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During the course of a study to determine the value of oral penicillin as a prophylactic measure for the prevention of streptococcal upper respiratory infections in rheumatic subjects, the following data on the absorption and excretion of this antibiotic were accumulated.

## PLAN OF STUDY

Oral penicillin* was administered over a twelve-month period to seventy rheumatic children ranging in age from 6 to 14 years, who were in the quiescent stage of the disease. A total of 100,000 units were given daily to each child in two doses. The optimum time of administration in relation to meals, variability of absorption in the same individual, and the relation of dosage to weight were studied. The penicillin tablets were all buffered with calcium carbonate, but no attempt was made to determine whether this buffer increased the absorption of the drug.

## METHOD

Samples of blood for the determination of penicillin levels were taken routinely thirty to sixty minutes after ingestion of the drug. Two-hour blood levels were studied in a small number of cases. Since penicillin is excreted in the urine almost entirely within the first six to eight hours,[16] close approximation of total urinary excretion was measured in eigh-hour urine specimens. Samples of urine were filtered through Seitz filters. Penicillin assays of serum and urine specimens were performed by a modification of the serial dilution method of Rammelkamp.[17] The test organism was a Group A hemolytic streptococcus C203 grown for eighteen hours in Todd-Hewitt broth. The Todd-Hewitt broth[18, 19] was modified as follows: beef was substituted for horse meat, 2 per cent neopeptone added, and sterilization accomplished by filtration through Berkefeld filters. An inoculum of 0.1 ml. of the freshly grown culture per 10 ml. of Todd-Hewitt broth was used, 0.5 ml. of which was pipetted into each of a series of ten tubes. Following the addition of 0.5 ml. of the unknown specimen to the first tube, serial dilutions were made. Similar dilutions of a standard penicillin solution beginning with a final concentration of 1 unit per milliliter in the first tube were included with each group of unknowns. After eighteen hours' incubation at 37° C., the end point was read as the lowest concentration of penicillin which would produce complete inhibition of growth. The maximum sensitivity with this method in the detection of penicillin in an unknown specimen varied between 0.031 and 0.0625 unit per milliliter.

## RESULTS

*A. 50,000 Units One Hour Before Breakfast.*—Following the administration of 50,000 units of oral penicillin on a fasting stomach one hour before breakfast, the penicillin concentration attained in specimens of blood drawn thirty to sixty minutes later was 0.20 unit per milliliter of serum, an average based on over 500 determinations. A smaller number of assays were done on

*The penicillin buffered with calcium carbonate (Ledercillin tablets) used throughout this study was generously donated by the Lederle Laboratories, Division, American Cyanamid Co.

samples drawn at accurately timed intervals of thirty, sixty and 120 minutes after administration of the drug. The average values of penicillin concentrations obtained at these intervals are presented in Fig. 1.

It will be noted that the maximum level of 0.29 unit per milliliter of serum was reached in thirty minutes, fell only slightly to 0.25 unit per milliliter at the end of sixty minutes, and then dropped to a low level of 0.04 unit per milliliter at the end of 120 minutes. No detectable levels were found beyond two hours.

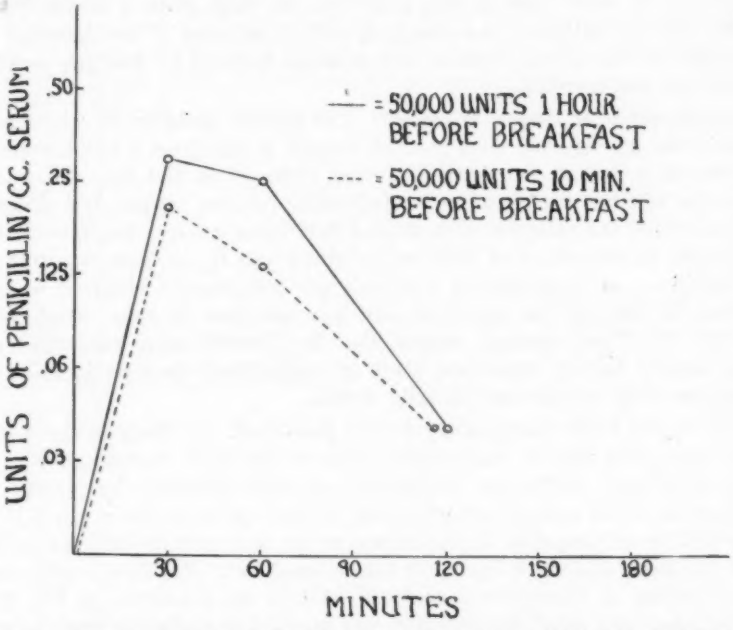


Fig. 1.—Penicillin serum levels following the administration of 50,000 units one hour before breakfast and ten minutes before breakfast. Individual figures are based on average of assays in twenty individuals.

The amount of penicillin excreted in eight-hour urine specimens following its administration in the fasting state ranged from 2 to 46 per cent of the ingested dose, with an average of 12 per cent in ten subjects so studied. Within the first 2 hours, 85 per cent of the penicillin was excreted and at the end of four hours about 99 per cent of the total penicillin excreted was present in the urine. Subsequent urine specimens contained only traces of penicillin.

*Variability in absorption.*—In the majority of children studied, namely fifty-five of the seventy, the serum levels obtained in determinations done on different days tended to remain remarkably constant. In the remaining fifteen children, on the other hand, marked day-to-day variations were observed. To exclude the possibility that in these fifteen individuals the maximum level was maintained for a shorter period of time and that slight differences in the time

that the blood sample was obtained might account for these variations, the time factor was accurately controlled. However, it was found that even with accurate timing the serum levels varied greatly on different days. A study of urinary excretion of penicillin in eight-hour urine specimens in these individuals indicated that the absorption of the drug from the duodenum varied from day to day, as indicated by the concentration of penicillin attained in the blood, and was directly reflected by the amount of urinary excretion. The findings obtained in one such individual are illustrated in Fig. 2.

It will be noted that in this individual on days when a serum level of 0.0625 unit per milliliter was obtained, only 3 per cent of the ingested dose was found in the urine, whereas with a serum level of 0.5 unit per milliliter, 26 per cent was excreted.

*Relationship of dosage to weight:* The seventy children in whom serum levels in the fasting state were studied, ranged in age from 6 to 14 years and represented a weight distribution varying from 49 to 136 lbs. In order to determine whether there was any relationship between weight and dosage of oral penicillin, the children were divided into three groups: the average penicillin level in the serum of children weighing 100 lb. or more was 0.16 unit per milliliter, as compared to 0.22 unit per milliliter in children weighing between 75 and 100 lb. and 0.24 unit per milliliter in those weighing less than 75 lb. These findings suggest that in children using equivalent doses under similar fasting conditions, there is a significant decrease in blood concentrations with an increase in body weight.

*B. 50,000 Units Immediately Before Breakfast.*—In forty subjects 50,000 units were given five to ten minutes prior to breakfast instead of one hour before breakfast. Following this method of administration, the average level obtained in blood samples drawn thirty to sixty minutes later was 0.15 unit per milliliter as compared to the average serum level of 0.20 unit per milliliter when the drug was given one hour before breakfast. The comparative serum concentrations at thirty, sixty, and 120 minutes are presented in Fig. 1. It will be noted that when the penicillin was ingested immediately prior to breakfast instead of one hour before breakfast, the concentrations attained in the serum at thirty- and sixty-minute intervals were somewhat lower although the general pattern of absorption was unchanged.

The average urinary excretion following ingestion just prior to breakfast was 8 per cent of the ingested dose as compared to the average of 12 per cent when the penicillin was administered one hour before breakfast.

*C. 50,000 Units Immediately After Breakfast.*—The ingestion of penicillin immediately after meals was investigated in two subjects. Both of these individuals had previously shown good serum levels when the drug was given before meals. However, in both subjects, when 50,000 units was ingested immediately after breakfast, the penicillin assay in blood samples drawn at thirty- and sixty-minute intervals revealed negligible concentrations. These findings were in complete agreement with those of other investigators, and this aspect of the study, therefore, was discontinued.

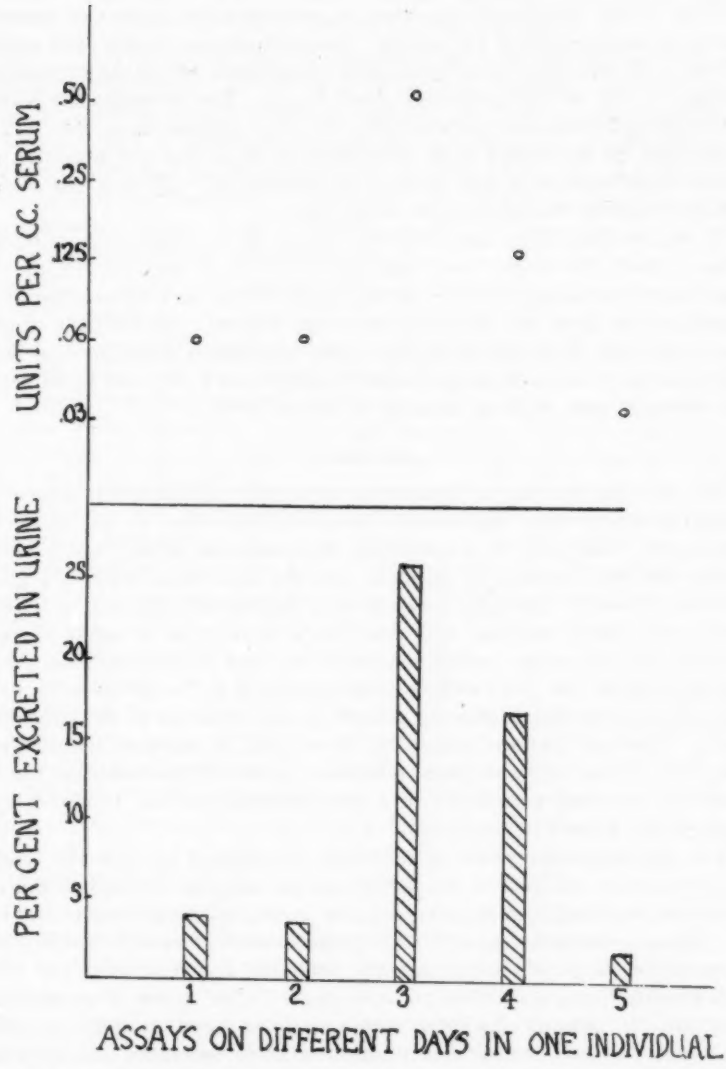


Fig. 2—Comparison of concentrations in sera thirty minutes after ingestion of drug and per cent of ingested dose excreted in eight-hour urine specimens in one individual who showed marked variations from day to day following administration of 50,000 units one hour before breakfast.

*D. 50,000 Units Immediately Before Supper.*—Since the ingestion of penicillin both one hour before and immediately prior to breakfast followed an overnight fast, it was considered important to investigate the degree of absorption following a shorter period of fasting. Accordingly, in twenty-four subjects, 50,000 units of penicillin was administered immediately before the evening meal, following a fast of approximately four hours. The average concentration attained in blood samples drawn thirty to sixty minutes later was 0.12 unit per milliliter as contrasted with an average of 0.15 unit per milliliter when the same dose was given just prior to breakfast and 0.20 unit per milliliter following ingestion one hour before breakfast.

*E. 50,000 Units Two and One-half Hours After Supper.*—In fifteen individuals, penicillin assays were determined in blood samples drawn thirty minutes after the evening dose. In a total of thirty such determinations, an assayable serum level was found in only one subject. However, a study of urinary excretion of penicillin in these same individuals following this method of administration revealed an excretion ranging from 2 per cent to 39 per cent of the ingested dose, with an average of 10 per cent.

## DISCUSSION

The general pattern of absorption of orally administered penicillin in children does not differ significantly from that described in adults by other investigators. The rise to a maximum concentration within thirty minutes indicates the rapid passage of the drug into the duodenum, where the greatest absorption occurs.[13] The peak serum level is maintained with only a slight fall for the next thirty minutes, and then drops rapidly to a barely detectable amount at the end of two hours. Since 85 per cent of the penicillin absorbed is excreted within the first two hours, the rapid fall in the serum concentration within the same period is directly related to the excretion of the drug by the kidneys. The fact that the remaining 15 per cent is excreted within the following two to four hours suggests that there is penicillin present in the blood beyond the two-hour period but in a concentration too low to detect by the method of assay used in this study.

The maximum absorption of penicillin, as reflected by both the height of the concentration attained in the serum and the amount excreted in the urine, occurs when the drug is administered in the fasting state one hour before breakfast. Using a constant dose of 50,000 units, somewhat lower but still satisfactory serum levels are obtained when the penicillin is given five to ten minutes before breakfast or immediately prior to supper after a fast of approximately four hours. If, however, the interval following the ingestion of food is reduced from four to two and one-half hours, only negligible concentrations are attained in the serum. Our findings in children are in accord with those of previous observers who have studied adults. Thus, Finland and associates,[15] using 50,000 unit doses in aqueous solution and with a variety of buffers, found that when penicillin was ingested thirty minutes after breakfast, the serum levels were irregular and unpredictable as compared to levels obtained when the drug was administered thirty minutes before breakfast. Similarly, Broh-Kahn and

Pedrick,[12] employing doses of 50,000 and 100,000 units without buffer, reported negligible serum levels one hour after breakfast, although the 100,000 unit dose given one hour before breakfast resulted in adequate levels.

The mechanism by which food interferes with the absorption of penicillin is not fully understood. According to McDermott and associates,[13] food tends to exert a buffering action on gastric acidity. In their opinion, food inhibits absorption mainly by mechanical means and by delaying the gastric emptying time. Broh-Kahn and Pedrick,[12] on the other hand, believe that gastric secretion is stimulated by the ingestion of food and hence greater destruction occurs in the postprandial state. These workers as well as György and associates[9] and Charney and associates[7] obtained satisfactory serum levels when penicillin buffered with trisodium citrate was administered after meals. In contrast to these investigators, in this study no evidence was obtained to suggest that buffering the penicillin with calcium carbonate increased absorption after meals. When the drug was ingested two and one-half hours after the evening meal, except for a single determination in one individual, no assayable serum levels were found. However, the average per cent of urinary excretion following this method of administration (10 per cent) compared favorably with that observed when the drug was ingested in the fasting state (12 per cent). These findings suggest that the presence of food in the stomach does not increase the destruction of penicillin to any great degree. It seems more likely the food delays gastric emptying time to the extent that the ingested penicillin reaches the duodenum in smaller concentrations, and assayable serum levels are not obtained.

The marked variability of blood concentrations observed in the same individual on different days which occurred in fifteen of the seventy children has been difficult to explain. If the amount of penicillin excreted in the urine is an approximate index of absorption,[13] studies of urinary excretion in these individuals indicate that the degree of absorption varies from day to day even in the fasting state. At the present time no adequate explanation of the variation in absorption observed in certain individuals is available. It seems possible that emotional factors even in the fasting state might influence the motility of the gastrointestinal tract.

Our results suggest that there is some relationship between dosage and weight. Using equivalent doses under similar fasting conditions, a significant decrease in blood concentration with increasing body weight was observed. The fact that most observers[12, 20] obtained only negligible blood levels with doses of 50,000 units in adults, in contrast to our findings in children, also indicates that there is a relationship between dosage and weight. Ross and associates,[8] on the other hand, comparing the blood levels of adults and children following ingestion of 100,000 units, found no difference between the penicillin concentrations in both age groups. Their findings suggest that the weight factor is probably less important when large doses are used.

In our opinion, oral administration of penicillin is a satisfactory procedure in children, provided that the drug is given as nearly as possible under fasting

conditions before meals, and if necessary during the night. If it is essential that the drug be given after periods of fasting of less than four hours, the dosage should probably be increased to compensate for the diminished absorption. Other factors which should be taken into consideration are the weight of the child and the occasional variability of absorption in certain individuals.

Although the children studied were all rheumatic subjects, they were in the quiescent stage of the disease. It therefore seems likely that similar results would be obtained in nonrheumatic individuals of the same age. No studies on the treatment of acute infections with oral penicillin were included.

## CONCLUSION

Satisfactory serum levels can be obtained in children following the oral administration of 50,000 units of penicillin, provided that the drug is given following a fast of at least four hours.

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# SELF-DEMAND FEEDING AND INDULGENCE IN EARLY INFANCY

GUSTAVE F. WEINFELD
HIGHLAND PARK, ILL.

MANY pediatricians believe that psychiatric principles have overemphasized liberty and permissiveness in the guidance of infants and children. They feel that "old-fashioned discipline" with its adult imposed restraints on behavior could be better relied upon to produce a generation of emotionally stable adults. There are countless examples in clinical experience, of parents and others who, under the guise of misinterpreted psychiatric concepts, have eschewed all repression, restraint, and inhibition in the guidance of their children. This is contrary to better psychiatric thought, which has recognized for a long time that overpermissiveness was not salutary for the older child. This does not apply, however, to the newborn and young infant, whose instincts and pleasures must be indulged and nourished if orderly emotional growth is to ensue.

Pediatrics is not, and never has been, exclusively a science of the cure and prevention of disease. The pediatrician is as much the pedagogue as is the academic teacher or the spiritual adviser. In fact, his influence is greater, because upon him falls the responsibility of directing the emotional development of the individual at a time in life when all the patterns of later life are being formed. This important role cannot be fulfilled without a knowledge of the nature of emotional growth trends and needs, any more than physical growth can be directed without a knowledge of the physiology of growth.

Nature did not intend that an infant should go through life receiving a constant stream of nutrition through his umbilical cord. Nor did she intend for him to be permanently spared the harsh impingement of physical forces by the protection of amniotic fluid and a uterine cloister. She endowed the normal infant with many capacities for his protection, the perception of his experiences, and the integration of them into a personality and character structure. The timing, dosage, and modification of the stimuli are important factors in the development of security and emotional growth.

During the early period of development, the mouth leads all other organs and sensory areas as a source of gratification, pleasure, and learning. Other forms of sensory perception are important, but they are secondary to the dominant oral needs. Feeding techniques and routines can provide the infant with the orderly learning of the pleasant uses of all his emerging senses and the gradual dawning that there can be safety and comfort in the ever increasing relinquishments and adaptations that society demands of him.

For some time after birth, the newborn infant functions with the same neuromuscular endowment which he had at his disposal during his intrauterine

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From the Department of Pediatrics, University of Illinois.

existence. Though many of the external and internal stimuli are now different, his reaction to them is qualitatively similar to that which it was before birth. Physical activity at this time is at an infracortical level and the responses are purely reflex in character. External stimuli, such as restraint or pain, or internal stimuli, such as produced by the intrinsic hunger contractions, all produce physiological tension which is released as crying and mass activity. Psychologically, at this age the infant is in a state of primary omnipotence. He is unaware of his environment as related to himself. He is aware only of his own tension and its release.

As maturation proceeds, physical activity gradually becomes more purposeful and voluntary. Cortical control has begun to emerge. Simple reflexes are progressively elaborated to complicated reactions under voluntary control. The state of primary omnipotence of the newborn is gradually replaced by an awareness of the relationships of self to environment. Certain aspects of growth, particularly the refinements of behavior, are being seriously influenced by the environment in which they occur. One may refer to this process in the physiological sense as conditioning, though in the broadly psychological sense it involves an adaptation to reality.

The instinctive need for food is physiologically expressed through the medium of hunger contractions. Pain occurs and tension accumulates as a direct result. It is well known that there is a great variability in the chronological rhythm with which these contractions occur. This rhythm differs from infant to infant and in the same infant at different times, but as growth proceeds, hunger occurs with progressive regularity. It is obvious, then, that any feeding schedule which provides for the presentation of food to the young infant at regular intervals is physiologically unsound. Such a schedule would not anticipate the natural rhythms of hunger and, therefore, food would be offered when it was not required, and withheld when it was demanded. This is not vitally important in the infant's nutritional economy, for he is dogged in his refusal to eat when not hungry and his capacities are surprising when he needs food.

The emotional aspects of hunger are also concerned with the irregularity with which hunger contractions occur. During the newborn period hunger contractions evoke tension and pain which is released by crying. The cry brings the mother and food. Gratification of an instinctual need occurs. At this point the mother and even the food is not recognized as part of the environment. When the feeding program is adjusted to the individual rhythm of the infant, hunger is always followed by gratification. When there is gratification, the sensory apparatus is released for ultimate recognition of an intermediary between hunger and gratification, the mother. When the feeding experience is satisfying and not frustrating, the infant comes to have confidence in the mother and his environment. He can depend upon the certain satisfaction of his instinctual needs and has learned to wait; he has made his first adaptation to reality. Equipped with a new attribute, confidence, all of his senses are expanded so that the rooting at the breast, handling of the breasts, the odor and taste of the milk, the sight of the mother, and the sound of her

voice, in fact all his perceptual senses, are constantly enlarged for the purpose of exploring reality. Under such circumstances, he is able to keep his instinctual need—hunger—in abeyance, deriving preliminary satisfaction through investigating the elements of his environment which he has learned through experience are the source or the means to his larger and more basic gratification, the relief of hunger.

There can be no doubt that the ideal feeding program should be based on the individual physiological and emotional needs of the infant. Such a program involves no fixed schedule whatsoever and the infant is offered food whenever he is hungry. The validity of this concept has been repeatedly established by numerous carefully controlled clinical experiments and observations. Some parents and pediatricians continue to resist the employment of self-demand schedules and refer to them disdainfully as a "newfangled idea." The idea, of course, is really as old as Nature, and was succinctly expressed by St. Marthe in 1584 in his Pediatrophia:

> "The hours of suckling it I do not fix
> Nature in that must guide the nursing sex
> When by its cries it calls you, do not spare
> Your labor, nor be loath your breasts to bare."

A few mothers find it difficult to employ what they consider a too liberal and revolutionary type of feeding and training regime. Usually they are women with a compulsive type of personality for whom orderliness and regularity are their sole protection against their own anxieties. For these parents, other less damaging outlets can be provided for their compulsions; these involve hours of bathing, types of clothing, exercise, room temperature, etc. These failing, a fixed schedule may be less thwarting to the infant's growth than the effects of a mother who is anxiety ridden by her anticipation of an unpredictable feeding time.

There is another type of mother who for one reason or another consciously or unconsciously rejects her infant. She eagerly accepts the authority of her pediatrician and sets about to fondle her baby and employ a self-demand schedule with grim determination as if these were inflexible tools in her hands. Such a mother may intellectually appreciate the rationale of the techniques involved, but emotionally she is unable to execute them. She fails because her frigidity, anxiety, or fear renders her incapable of creating infantile gratification and security regardless of techniques employed.

Many parents believe that feeding the baby when he is hungry, allowing him to suck whenever he wishes, fondling or rocking him, and even speaking softly to him will stunt the development of his moral strength. They feel that because of such permissiveness and indulgence he will never learn to keep his impulses in check or hold his needs for sensory pleasure in abeyance. There is, of course, no danger that this will occur. The newborn is not endowed with any faculties for consciously testing the interplay of his instinctual wishes with the forces of reality. It requires many months before the infant is capable of deciding whether his immediate instinctual gratification is more important than

a delayed but greater gain. An early basic security as acquired through past feeding experiences, and sensory satisfactions will enable him to keep his instincts and hunger in check and to wait for the mother whom he has learned has never failed him. This capacity to weigh the demands of the instincts and pleasure needs is slow in growing and is acquired only by a repetition of safe and pleasant experiences over a long period of time. Unsatisfied hunger in a 6-week-old infant produces a rage reaction. A 3-year-old whose early instinctual life has been gratified feels secure; he knows that he is loved and has learned through experience that his patience will be fully rewarded by a mother whose dependability has never failed him. In fact, his compliance is a reciprocal gift for her goodness and love. The less fortunate 3-year-old who is not genuinely loved or who, because of lack of parental insight, has been denied the gratification of his elementary needs, will not be able to wait. He will not give patience, consideration, compliance, and love because he has received none. He has not been able to progress emotionally much beyond the 6-week-old infant whose only reaction to frustration is rage.

## CONCLUSION

The security which arises from the gratification of primitive instinctual needs by self-demand feeding regimes and other forms of sensory indulgence is an important asset for later emotional learning experiences; the older infant who feels secure can then begin to accept the training programs involving denial and frustration.

# METHOD OF HOLDING THE HEAD FOR SCALP VEIN TRANSFUSIONS AND RESTRAINING THE LEGS FOR INTRAVENOUS THERAPY

PETER H. SPOHN, M.D., AND TAYLOR STATTEN, M.D.
TORONTO, ONT.

AN IMPORTANT feature in successfully carrying out many pediatric procedures is to have the child securely restrained. This is particularly the case when attempting to keep a needle in a superficial scalp vein, or in cutting down on a vein for continuous intravenous therapy. Two types of apparatus have been devised at the Hospital for Sick Children which have proved useful for holding the head and lower limbs of the patient while carrying out these procedures.

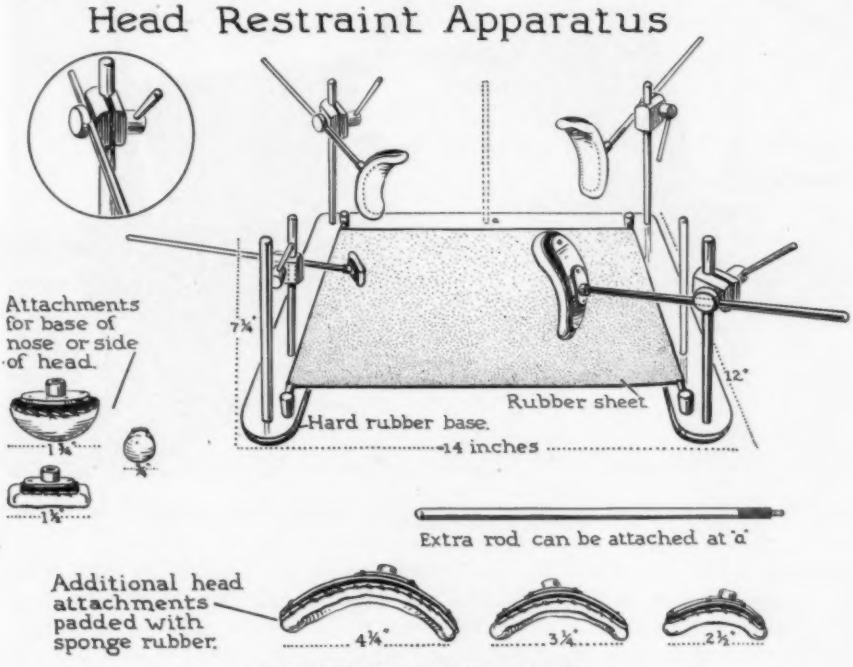


Fig. 1.—The head restraint apparatus.

Formerly it was necessary to have the child held by a nurse while performing a scalp vein transfusion. This is a very tedious task for the nurse and experience has shown that the head can be held more securely in the head re-

From the Hospital for Sick Children and the Department of Paediatrics, University of Toronto, under the direction of Alan Brown, M.D., F.R.C.P. (London).

straint apparatus. The child is also less distressed when held by this apparatus and the transfusion is administered with less difficulty.

The head restraint apparatus is illustrated in Fig. 1. The apparatus consists of four 7-inch posts and two 6-inch posts mounted on a metal base. There is also an extra post which can be attached at position *a*. Four arms are clamped to the post by an adjustable clamp as shown in the insert, Fig. 1. Various sized head pieces can be screwed to the ends of the arms to fit any shaped head. The arms of the apparatus are then adjusted to the various posts,

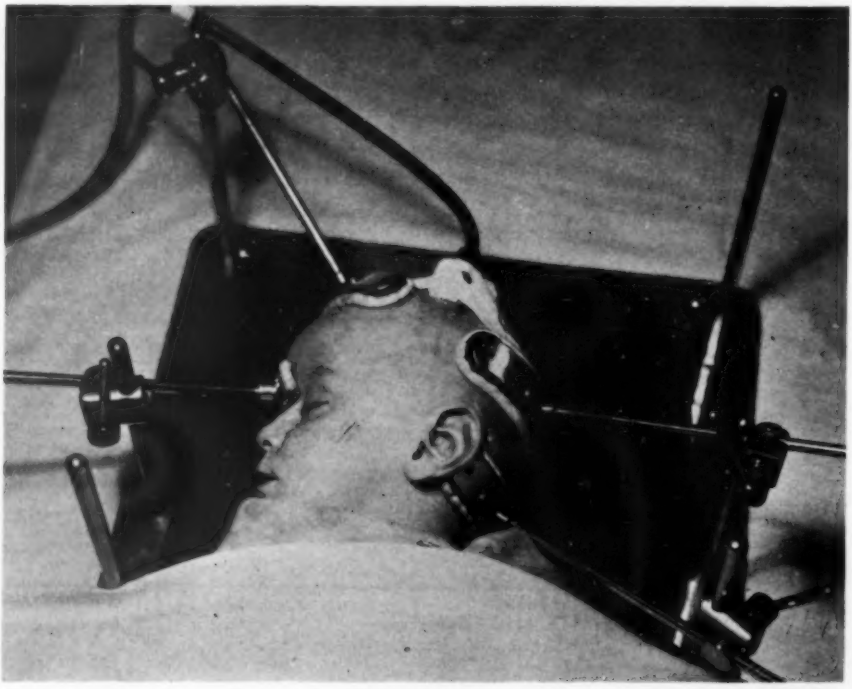

Fig. 2.—Illustration of the method of holding the head in the lateral position during a scalp vein transfusion.

depending on the size and shape of the child's head. One arm is fitted with a nose-piece which is applied to the base of the nose. The head rests on a suspended rubber sheet, and the head and nosepieces are adjusted as demonstrated in Fig. 2. It is important to have the child well restrained and to fasten the shoulders and body to the table by a sheet as illustrated in Fig. 2. As well as the lateral position, the head can be held securely in the upright position by adjusting the arms from the 6-inch posts contacting each side of the head in the parietal region.

The leg restraint apparatus allows the lower limbs to be held securely while the intravenous is being started and while fluids are being administered. This

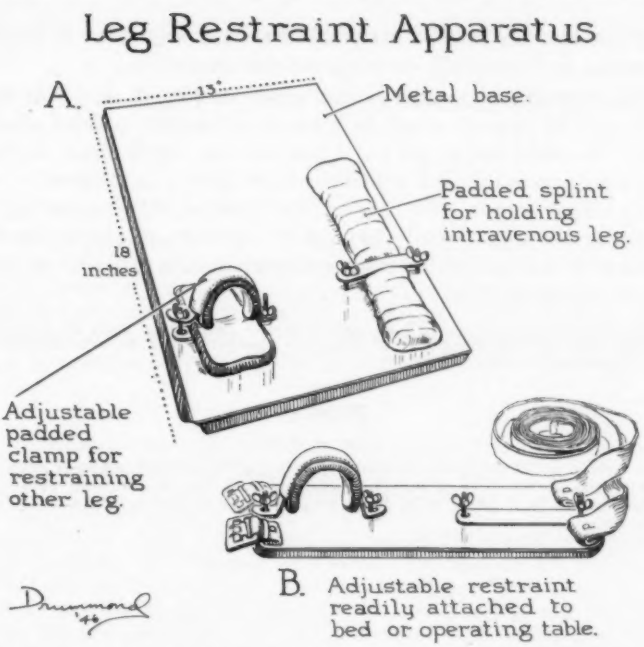


**Fig. 3.**—*A*, The leg restraint apparatus. *B*, The adjustable leg restraint apparatus.

**Fig. 4.**—Illustration of the method of holding the legs while giving continuous intravenous fluids.

method of restraint has saved considerable time formerly spent in fastening the legs by various methods while carrying out this procedure.

The leg restraint apparatus is illustrated in Fig. 3, *A*. This apparatus consists of a metal base on which is fitted an adjustable, padded clamp and a metal bar. The child lies on the metal base and one leg is placed in the clamp. The other leg, on which the intravenous is to be started, is strapped to a padded splint with adhesive tape. The bar is then inserted between the leg and the splint and attached to the base by two screws, as shown in Fig. 4. In Fig. 3, *B*, is illustrated the adjustable leg restraint which can be attached by straps to a cot or to an operating table.

This apparatus was originally constructed in the Orthopedic Shop of the Hospital for Sick Children, Toronto. Information regarding this apparatus may be obtained through J. F. Hartz and Company, Toronto, Ontario.

## Erratum

In the article entitled, "Acute Glomerulonephritis" by Burke and Ross in the February, 1947, issue of the JOURNAL OF PEDIATRICS, on page 170, line 6, the dose of Digifolin was stated to be one cat unit per 100 lb. body weight. This should have read one cat unit per 10 lb. body weight.

# Case Reports

## IDIOPATHIC DILATATION OF THE COMMON BILE DUCT IN CHILDHOOD

WESTON M. KELSEY, M.D.
WINSTON-SALEM, N. C.

IDIOPATHIC dilatation of the common bile duct is rare. Seventy-one case reports have been found, of which only two articles are in the American pediatric literature.[1, 2] Many cases are reported in journals which are not available, such as forty-two cases reported from Japan. This condition has rarely been considered preoperatively. The frequency with which the condition is overlooked is probably a result of lack of publicity, as the symptoms and signs are typical.

### CASE REPORT

B. F., a white female, aged 4 years, was admitted to the Pediatric Service of the North Carolina Baptist Hospital on Aug. 9, 1946.

*Family History.*—The father was killed in action, the mother was living, and there were no other siblings.

*Present Illness.*—The patient had an uneventful neonatal period without physiologic jaundice. At the age of 3 weeks, she had an episode of jaundice which lasted one week and subsided without therapy. She remained well until the age of 15 months, at which time there was an attack of vomiting and fever followed by marked jaundice. The urine became dark and the stools acholic. The patient's status remained unchanged for a period of eight months, at which time choline was given. Shortly after this the patient improved rapidly. This was reported in the literature as a cure of hepatitis with choline.[3] At the age of 26 months, jaundice developed again. From that time until the onset of the present attack, the patient had many minor illnesses with mild jaundice, dark urine, and light-colored stools. The last episode began two months prior to admission. During this period she complained of right upper abdominal pain for the first time. The jaundice, dark urine, and acholic stools persisted.

During these illnesses the patient had been seen elsewhere by three competent pediatricians, who felt she had recurrent hepatitis with cirrhosis. She was referred to the North Carolina Baptist Hospital for further study.

*Physical Examination.*—Revealed a well-developed and well-nourished child, who was not acutely ill. The temperature, pulse, respirations, and blood pressure were normal. The height was 42½ inches; weight, 37½ pounds. The skin and sclerae were markedly icteric. The abdomen was protuberant. The liver was palpable 8 cm. below the right costal margin. The spleen was palpable 2 cm. below the left costal margin. No tumor mass could be felt in the abdomen.

*Laboratory Findings.*—Urinalysis: bilirubin and bile were present, and urobilinogen was present in minimal amounts. Blood: 11 Gm. hemoglobin; red blood cells, 3,490,000; white blood cells, 7,350; polymorphonuclears, 58 per cent; lymphocytes, 40 per cent; eosinophiles, 2 per cent. Fragility test: hemolysis began at 0.40 per cent and ended at 0.32 per cent; control began at 0.46 per cent and ended at 0.38 per cent. Bilirubin was 11.5 mg. per cent. Serum cholesterol was 446 mg. per cent. Total serum proteins were 6 Gm. per cent. Stools were gray. Test for bile was weakly positive. Prothrombin time was 16.4 seconds. Control was 16.5 seconds.

From the Department of Pediatrics, Bowman Gray School of Medicine of Wake Forest College, and the North Carolina Baptist Hospital.

It was felt that she had an obstructive lesion in the biliary tract, probably a choledochus cyst.

*Operation.*—Performed Aug. 17, 1946, by Dr. Felda Hightower.

The gall bladder was distended. A large cystic mass was found below the neck of the gall bladder. Exploration revealed that this was the dilated common duct. No calculi were found. An anastomosis was made between the common duct and the second portion of the duodenum. Culture from the gall bladder was sterile.

*Postoperative Course.*—The course was uneventful except for two days of unexplained fever. The icterus cleared rapidly. The liver was palpable 3 cm. below the costal margin when she was discharged on the fifteenth postoperative day.

*Laboratory Findings on Discharge.*—Serum bilirubin was 2.3 mg. per cent; cholesterol, 204 mg. per cent; total serum protein, 6.5 Gm. per cent; bile test on the stool, strongly positive.

Five months after discharge the parents wrote that the patient had had no recurrence of symptoms and was well.

## COMMENTS

The only unusual feature in this case was the absence of a palpable tumor. The coincidental improvement when choline was given erroneously supported the diagnosis of hepatitis, and delayed exploration at least twenty months.

## DISCUSSION

The numerous case reports since the excellent review of Gross[1] have added no new information concerning the etiologic factors in this condition. The etiology and the mechanism of the recurrent jaundice remain obscure. The common triad of symptoms is jaundice, right upper quadrant pain in the abdomen, and a palpable tumor. This triad has been present in about 60 per cent of the cases. About 85 per cent of the patients have been females. The mortality in those cases in which the diagnosis had been considered preoperatively was approximately half that in the remainder.[4] Few long-term postoperative observations have been made in these patients. McWhorter[5] reported a woman who had a normally functioning biliary system thirteen years after operation. Cirrhosis[6] and biliary calculi[7] have been reported as complications. There are no reports of spontaneous cure.

## SUMMARY

A case of idiopathic dilatation of the common duct in a 4-year-old girl with successful operation is reported.

The importance of considering the condition in recurrent jaundice in childhood is emphasized.

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# IMPETIGO NEONATORUM CONGENITA

## Five Cases in Siblings

Abraham Gilner, M.D., and Irving Nelson, M.D.
Brooklyn, N. Y.

THE congenital form of pemphigus neonatorum, or impetigo neonatorum congenita, is a rather rare condition. In 1908, Labardt and Wallart[1] reviewed thirteen cases going as far back as 1794, and added three cases of their own. Grosz[2] reported one case of his own, and recalled a similar case of Peters, in 1902. Since that time, only a few cases have been added to the literature. Reed[3] brought the attention of American dermatologists to the possibility of congenital pemphigus in 1928 when he reported eleven cases, nine of which he had personally observed. Freeman[4] reported two cases in 1931. Wise, in discussing Freeman's paper, stated that he had seen one case of congenital pemphigus. In 1934, Rucker[5] reported neonatal deaths in two siblings from impetigo neonatorum congenita. Weston[6] reported one fatal case in 1936, while Daniel and Ayer[7] observed another case with a similar termination in 1937. DeLee, in a personal communication to Daniel and Ayer, stated that up to July, 1936, he had known of only one case, in which a woman had delivered two babies, both born with pemphigus. This is the report of one of five cases in siblings, in all of whom a diagnosis of congenital pemphigus neonatorum was made.

### CASE REPORT

Mrs. L., 34 years old, colored, a nonclinic patient, delivered spontaneously a 9-pound girl on Aug. 1, 1946. Immediately after delivery it was noticed that there was marked maceration of the infant's skin. Desquamation was most marked around the posterior portion of the neck, with extension around to the angles of the jaw. A small portion of the upper anterior chest, extensor surface of the right forearm, and, to a lesser extent, the palms and soles, were also involved. A few scattered milia were found on the lower jaw. Several vesicles about 2½ mm. in diameter were scattered on the left anterior chest and lower back. The palms and soles were free of any vesicles or bullae. No lesions could be found around the anus, in the mouth, or around the eyes. The cry, color, and respiration were good. Heart and lungs were normal, while the liver and spleen were not palpable.

Both mother and baby were immediately placed in isolation. A dark-field examination of the base of one of the vesicles was done the day of delivery and revealed no organisms. Fluid was aspirated the day following delivery from another vesicle and cultured. *Staphylococcus aureus* was grown. A blood culture taken the same day was sterile. A blood count revealed: hemoglobin, 112 per cent; red blood cells, 5,075,000 per cubic millimeter; differential—stab cells, 11 per cent; polymorphonuclears, 41 per cent; lymphocytes, 38 per cent; monocytes, 2 per cent; and eosinophiles, 8 per cent. The cord Wassermann and Kline were both negative. Another vesicle was aspirated on August 3, the fluid cultured, and *Staph. aureus* again grown out. A blood count done on August 21 showed: hemoglobin, 96 per cent; red blood cells, 4,760,000 per cubic millimeter; white blood cells, 12,000 per cubic millimeter; polymorphonuclears, 32 per cent; lymphocytes, 66 per cent; and eosinophiles, 2 per cent. An x-ray of the long bones, wrists, and ankles taken on August 1 was normal.

From the Service of Dr. J. Monfort, Department of Pediatrics, Cumberland Hospital. Brooklyn, T. B. Givan, M.D., Director of Service.

During her stay in the hospital the baby's general health remained very good. The condition of the skin was followed very closely. It was noted that new crops of vesicles would appear at about two- to four-day intervals. These vesicles were found scattered over both upper and lower extremities, the back, in the axillae, behind the ears, and in the inguinal folds (Fig. 1). The lower anterior chest and abdomen were spared. When first seen, the vesicles contained clear fluid. These would then rupture within the following twenty-four to forty-eight hours with a localized desquamation subsequently occurring. Oozing and maceration of the skin was quite marked in the folds. Considerable crusting of the scalp was also present. By the time the patient was discharged on Sept. 5, 1946, there was present, however, only a slight crusting of the scalp and a pigmentation of the upper and lower extremities. The discharge diagnosis was pemphigus neonatorum congenitus.

Fig. 1.—Photograph of first patient, taken on fifth day, showing involvement of the scalp, forehead, upper chest, and both upper and lower extremities.

Local medications used at various times were 5 per cent sulfathiazole ointment, zinc oxide, and boric acid ointment. At one time penicillin, 10,000 units, was given intramuscularly every three hours for a period of three days.

The following history and information were obtained from the mother, and the records of the Cumberland and another local hospital.

In 1934, the patient delivered a normal male at the Cumberland Hospital after having attended the prenatal clinic. Her blood Wassermann at the time was negative. In 1935, a girl was delivered at the Cumberland Hospital, and the Wassermann at the time was negative.

On Aug. 31, 1936, the mother attended the prenatal clinic and was told that she had "what looks like a healed secondary skin eruption." The blood Wassermann done with the cholesterin antigen was one plus, while with the alcohol antigen it was negative. The diagnostic and exclusion blood Kline were both one plus. The mother was then referred to an antisyphilitic therapy clinic. She received weekly intravenous and intramuscular injections for one year and was then discharged as cured.

On Oct. 3, 1936, a full-term female baby was delivered at the Cumberland Hospital. "Baby had vesicles scattered over its body. Lesions developed more characteristically, becoming generalized pemphigus." X-ray of the long bones was negative for osteochondritis. The baby's blood Wassermann was negative on Oct. 24, 1936, while that of the mother was one plus. The baby was followed in the clinic up until Jan. 6, 1940, never showing any signs of congenital syphilis. During that time, six blood Wassermann tests were all negative.

During 1937 and again in 1938 the mother had two spontaneous abortions at three months each. She was treated at a local hospital. Blood Wassermanns were negative both times.

Since 1938, the mother had been subject to repeated attacks of outcroppings of small blisters around her neck. Regardless of what remedy she would use, the blisters would eventually crust, dry, and disappear. These attacks had occurred with increasing frequency with each succeeding pregnancy. In addition, the attacks were also worse for a period of about four to five weeks following parturition. She had attended several skin clinics but had never been told what was wrong with her.

On Aug. 7, 1939, the patient delivered an 8-pound, 6-ounce male "apparently normal except for maceration of skin over fingers and toes and circinate erythematous skin lesions over the face with raised maculopapular margins and isolated vesicles, these being dark red, and the surrounding skin macerated." The cord Wassermann was negative as was the blood Wassermann done on March 16, 1941. X-ray of long bones on Jan. 15, 1940, was also negative. After discharge from the hospital, the baby was treated in the skin clinic for seborrhea of the scalp, which condition eventually cleared up.

Jan. 7, 1943, saw the birth of a girl at another local hospital. Irregular outlined bullae were present on the baby's body at birth. Small pustules were also present in the scalp. A smear taken from one of the pustules showed degenerated white cells and gram-positive diplococci. The blood Wassermann of the baby was negative while that of the mother was one plus. The infant was discharged with a diagnosis of impetigo neonatorum. The skin lesions eventually cleared up at about 2 months of age.

On March 5, 1944, the patient delivered a girl at the Cumberland Hospital. At about one week prior to delivery the mother had received one intravenous and one intramuscular injection at an antisyphilitic therapy clinic. At the time of delivery it was noticed that the baby's skin over the forehead was macerated and that many small vesicles could be found over the body. From the time of delivery up until the day the baby was discharged, March 28, 1944, the skin lesions went through the same stages of vesicle formation, drying, and residual hyperpigmentation as the previously mentioned cases. Two dark-field examinations of fluid from the vesicles, cord Wassermann, another blood Wassermann done at 4 weeks of age, and x-ray of long bones were all negative. *Staph. albus* and *aureus* were grown from the aspirated vesicle fluid.

Examination of the mother revealed "a diffuse skin lesion with eroded corium and shiny pearly gray base varying in size from 0.5 to 9 cm. in diameter beneath the breasts and in the inguinal and lower abdominal areas. Some have dried crusts on them." Diagnosis was intertrigo (dermatitis factitia).

A blood Wassermann of the mother was negative in May, 1945.

## DISCUSSION

The congenital form of pemphigus neonatorum is, as a rule, a benign disease. It does not affect the general condition, and the vesicles or bullae disappear and heal within a few weeks, never to recur, and leaving no scars. The cases we reported followed this pattern very closely.

The problem of etiology remains to be solved. When the infants were delivered a tentative diagnosis of congenital syphilis was made in all five cases. Suspicion was especially high in the infant which had been born during the time the mother was receiving antisyphilitic therapy. The negative x-ray of long bones, negative dark-field examination, and persistently negative blood Wassermanns over a period of three years ruled out the possibility of congenital syphilis. These findings, in general, also held true in the other four cases.

Both streptococci and staphylococci have also been assigned the role as the causative agent. Two of our cases showed staphylococci while another showed gram-positive diplococci. The latter may have eventually grown out to have been either a staphylococcus or streptococcus.

The mothers of the cases reported in the literature were all healthy and manifested no external lesions. We feel that in our case there must have existed a close relationship between the skin lesions of the mother and those of the siblings. The mother had been subject to attacks of outeroppings of small blisters around her neck which would eventually dry, crust, and disappear, a course closely followed by the skin lesions of her children. These attacks had become more severe with each succeeding pregnancy. It is highly possible that there might have been a transmission of the infection from the mother by transplacental or hematogenic means. Unfortunately, no cultures or smears of the mother's cutaneous lesions had ever been done.

## SUMMARY

1. Five cases of pemphigus neonatorum congenitus are added to the previously reported cases.

2. In only two previous instances has the disease been definitely reported as occurring in siblings. All five of our cases were in siblings.

3. The similarity of skin lesions suggests a transplacental or hematogenic spread of the infection from the mother to the infants.

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# CONGENITAL DESMOID OF THE ABDOMINAL WALL

DAVID DEAN BROCKMAN, M.D.
DURHAM, N. C.

DESMOIDS are among the less commonly occurring connective tissue tumors; the congenital desmoid is extremely rare. Pack and Ehrlich[1] reported an incidence of 0.03 per cent in their series from the Memorial Hospital. In 4,660 autopsies at Duke Hospital two instances of this neoplasm were found, or 0.043 per cent. In the surgical pathologic series of 114,778 specimens, ten cases, or 0.0087 per cent, were indexed. The case reported in this paper represents the only congenital desmoid in either series.

Pfeiffer[2] (1904) collected eight cases of childhood desmoid of the abdominal wall at the Bruns Clinic, two of which are definitely congenital, one of them having been described previously (1896) by Kramer.[3] The ninth case was reported by Sujoy and Maggi.[4] Grieg[5] (1915) described what he thought to be a congenital desmoid, but the tumor was first noted at the age of 8 months. The following is believed to represent the third unquestionable congenital desmoid of the abdominal wall to be described.

## REPORT OF CASE

The patient was a 4-week-old Negro female infant born without complication following full-term gestation. At birth a midline tumor was noted extending from the pubis to within one centimeter of the umbilicus; the skin at one point over the mass was ulcerated and bled somewhat for several days. Despite the presence of the tumor, the infant gained weight and grew at a normal rate. The family history contained no pertinent information. The physical examination was negative except for the discovery of the tumor. This measured 15 × 8 cm.; it was solid, irregular in outline, firm, symmetrically disposed, fixed to the underlying tissues in the midline, attached to the overlying skin, and did not transmit light or emit peristaltic sounds. It extended from the mons veneris to 1 cm. below the umbilicus. The skin overlying the most prominent portion was thin, dry, discolored, and at one point there was an ulcer 2 cm. in diameter. Distally, the mass impinged upon the labia majora. The urethral meatus was found at the inferior pole of the tumor.

Laboratory studies of blood and urine revealed no abnormal findings. Roentgenograms showed the tumor to be of uniform density and not connected with adjacent bony structures. The hospital course was uneventful until surgical removal of the tumor was undertaken. The tumor was found to infiltrate the rectus muscles and their sheaths, but did not extend to the peritoneum. It was removed, but the patient did not survive the necessarily extensive operative procedure.

*Autopsy (19 Hours After Death).*—There was a T-shaped midline, sutured, surgical incision below the umbilicus. The longitudinal arm was 5 cm., and the suprapubic transverse arm 7 cm. long. The external genitalia exhibited moderate edema. The remainder of the postmortem examination was without abnormal findings.

The surgical specimen measured approximately 20 × 8 × 4 cm.; the cut surfaces were smooth, firm, trabeculated, white, and glistening. The skin was intimately adherent to the tumor, and the rectus sheaths blended imperceptibly with the neoplasm. The rectus muscles were replaced except at the superior pole of the tumor. No definite capsule could be made out.

Microscopically the neoplasm was found to be very cellular. Near the center of the mass the cells were identical with adult fibrocytes. The elongated cells formed strands and bundles which interlaced in all directions after the manner of the common uterine fibromyoma. Extension of the neoplastic cells was found well into the corium of the overlying skin. At the periphery of the tumor

From the Department of Pathology, Duke University School of Medicine.

Fig. 1.—Ante-mortem photograph of infant showing tumor in the abdominal wall.

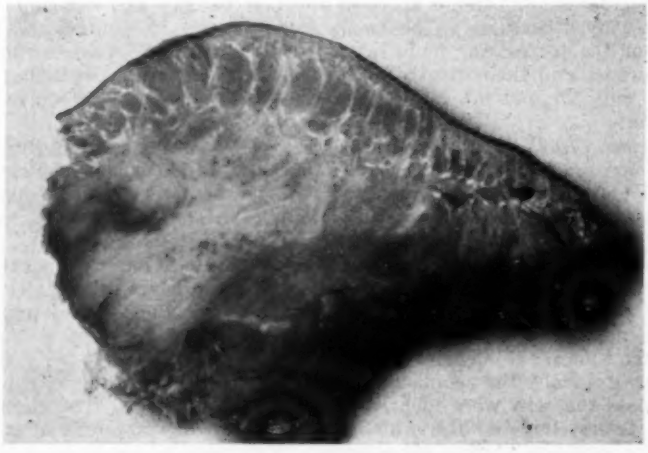

Fig. 2.—Gross appearance. The neoplasm presents a smooth, firm, trabeculated, white, and glistening cut surface.

fibroblast-like cells infiltrated the skeletal muscle. The cytoplasm of the skeletal muscle fibers in small areas were found to be undergoing degenerative changes. Multinucleated giant cells with peripheral or polar nuclei were present. Special stains for fibrous tissue, neurofibrils, myelin, and reticulum merely confirmed the fibrous nature of the tumor.

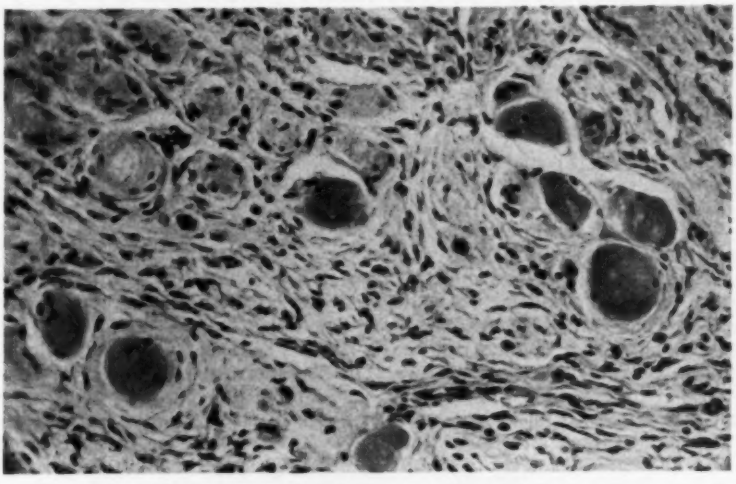

Fig. 3.—Photomicrograph showing the formation of multinucleated masses from degenerating striated muscle. (×250.)

## COMMENT

Since the first description of the desmoid tumor by MacFarlene[6] (1832) numerous ideas have been advanced as having etiologic significance; among these are pregnancy, trauma, and operation scars. Geschickter,[7] investigating the hormonal influence in desmoids, has recovered 13,000 units per kilogram of gonadotropic hormone by bio-assay. No estrin was obtained. None of these ideas has clarified the congenital desmoid, which unquestionably arises at a relatively early embryonic age. Of the several cases of this tumor reported in children, all but two were discovered two months to two years postnatally. It is more logical to reserve the term "congenital" desmoid for those instances in which the tumor was noted at birth.

The attendant difficulties and dangers of surgical and obstetrical management of the congenital desmoid are apparent.

## SUMMARY

A case is presented which constitutes the third reported unequivocal instance of the congenital desmoid, i.e., discovered at birth. Ten cases of abdominal wall desmoid in infants and children have been recorded in the literature, only two of which were noted at birth. The morphologic and histologic characteristics are reviewed.

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# LIPOCHONDRODYSTROPHY (GARGOYLISM)

M. E. Lahey, M.D., R.D. Lomas, M.D., and T. C. Worth, M.D.
Houston, Texas

LIPOCHONDRODYSTROPHY is a rare familial disease of undetermined etiology. In its fully developed state the clinical appearance is that of a retarded, disproportionate dwarf with gargoylelike facies, corneal opacities, kyphosis, and a protuberant abdomen. In addition to the name used here,[1] the disease is variously known as Hurler's Syndrome,[2] gargoylism,[3] and dysostosis multiplex.

While the appearance at birth may be normal, some signs of the disease are almost always evident in the second six months of life. The earliest of these signs may be the corneal opacities, enlargement of the head, dorsolumbar kyphosis, developmental failures, or a combination of these. As time passes, and usually by the age of 4 years, the disease in its entirety is manifest. Dwarfism is apparent, and the trunk seems too short for the legs. The head is large, may be deformed, and the neck is short. The external ears may be displaced caudally. The eyes are wide-set, and may show proptosis and corneal opacities. There is a depressed nasal bridge and frequently a chronic discharge. The lips and tongue are thick and the teeth widely separated and poorly formed. There may be flaring of the costal margins, blending into the rounded protuberant abdomen. Hepatosplenomegaly and an umbilical hernia are frequent. The kyphosis may be marked or absent. Flexion deformities of various joints are usually apparent, with more or less typical thick, broad, "claw" hands. Enlargement of the wrists is sometimes noted. There is impairment of the intelligence in most cases. This description of the clinical features may be found in various reference works on this disease.

Roentgen-ray studies of the skeleton reveal some or all of these abnormalities: (a) large misshapen skull with an elongated, shallow sella turcica; (b) thickened sternal ends of the clavicles; (c) kyphosis involving the twelfth dorsal, and first or second lumbar vertebrae; the anterosuperior portions of the vertebrae involved in the kyphosis may be destroyed, resulting in a hooklike projection from the anteroinferior portion; (d) lower ribs constricted at their necks and broad beyond, giving a "ribbonlike" appearance; (e) shallow glenoid and acetabular fossae; (f) heads of the humeri tilted medially; (g) shafts of humeri swollen with tapering toward their ends: (h) coxa valga deformity; (i) distal epiphyseal lines of radius and ulna tilted toward one another; (j) retardation of "bone age;" (k) metacarpals and phalanges broad, short, pointed, with widened medullary cavities and coarse trabeculations throughout.[4-6]

Routine laboratory and blood chemistry studies are usually within normal limits. However, a microcytic, hypochromic anemia may be present. Attention has been called to the presence of large granules and occasionally vacuoles in the cytoplasm of the polymorphonuclear leucocytes.[7] Hypercholesterolemia and impaired liver function have been reported.[8] Air ventriculography may show dilatation of ventricles; at least one electro-encephalogram was normal.[8]

The prognosis is poor and treatment is almost always ineffective.[1]

The disease is said to show no racial limitations and there have been reports in various nationalities. While no painstaking search of the literature was made, this is the first case, to our knowledge, to be reported in a Negro.

From the Department of Pediatrics, Baylor University College of Medicine, the Hermann Hospital, and the Junior League Children's Clinic.

CASE REPORT

P. A. L., Negro, female, was breech-born at full term after a normal labor, and weighed 2,700 Gm. No abnormalities were noted at birth. The parents were said to have been healthy and were unrelated. There was no history of similar disease in relatives.

The patient sat alone at 8 months, cut her first teeth at 12 months, and was able to "pull-up" at 19 months. The words "mama" and "daddy" were first spoken at 17 months.

Fig. 1A.—Front view showing characteristic clinical appearance. Note the claw hand.

Soon after birth "noisy" respirations and a watery nasal discharge were noted. A "lump in the back" was first noted at about 1 year, and haziness of the eyes at about 2 years. There was always fecal and urinary incontinence. Hearing seemed unimpaired.

At the age of 19 months the diagnoses of rickets, scurvy, and anemia were made, for which the patient received the appropriate vitamins and a blood transfusion.

One year later, i.e., at the age of 2 years, 7 months, she was admitted to the Hermann Hospital for diagnostic studies. At the time the patient could sit alone, crawl, pull up to a standing position but not walk. She was quite irritable and resisted examination vehemently. Speech was limited to the words "mama" and "daddy."

The general appearance was as shown in Fig. 1, a pot-bellied child with large head, grotesque facies, and dorsolumbar kyphosis. The fronto-occipital circumference was 51.5 cm., height 79 cm., weight 11.8 kg.; the center of the body was at the umbilicus. There was fine black hair over the shoulders. The nasal bridge was depressed and there was a profuse mucopurulent nasal discharge.

Fig. 1*B*.—Profile view showing characteristic clinical appearance.

The eyes were wide-set and with slight proptosis. There was milky-white opacity of both corneas with no vascular infiltration. Because of the patient's uncooperativeness the slit lamp was not used. It was the ophthalmologist's opinion that probably only the superficial portion of the cornea was involved. The lips were thick. Nine teeth were widely spaced and peg-shaped. The neck was short and held in slight extension. The ears were displaced caudally. The thoracic cage flared at the costal margins. The heart sounds were forceful, but no murmurs were heard. There was diastasis recti and a small umbilical hernia. The liver edge was firm and extended 6 cm. below the right costal margin. The spleen was not felt. There was limitation of abduction at both shoulders and of extension at the elbows and knees. The hands were held in slight palmar flexion and fingers were flexed, as shown in Fig. 1, *A*.

Laboratory data revealed a hypochromic microcytic anemia. No abnormal granules or vauoles were seen in the cytoplasm of the polymorphonuclear leucocytes, as Reilly suggested.[7] No bone marrow studies were done. Examination of urine and stools was negative. Wassermann and tuberculin tests were nega-

tive. Blood cholesterol was slightly elevated (278 mg. per cent). X-rays of the skeleton revealed almost all of the previously listed abnormalities, as depicted in Figs. 2 to 6. The skull is not shown, but it did reveal symmetrical enlargement with a suggestion of elongation of the sella turcica. The bone age was not retarded and the sternal ends of the clavicles were not abnormal.

Fig. 2.—Kyphosis and a hooklike projection from the anteroinferior portion of second lumbar vertebra are shown.

## DISCUSSION

From the foregoing review and case report it is obvious that a combination of seemingly unassociated features is present in this disease. It is this fact which makes for confusion so far as the etiology is concerned.

Some contend that a defective germ plasm accounts for the disturbance in endochondral growth; the nonskeletal features are said to result from a deranged lipoid metabolism, causing this disease to be classified as the "fifth lipoidosis" along with Niemann-Pick, Tay-Sachs, Hand-Schuller-Christian, and Gaucher's diseases. It is stated that pathologic studies substantiate this argument.[9-11]

Engel[12] applying the works of Bagg and of Bonnevie, has proposed a very interesting theory of the etiology of this and related diseases showing multiple

Fig. 3.—Broad, "ribbonlike" lower ribs are shown.

Fig. 4.—A portion of the upper extremity showing the swollen shaft of the humerus and the medial tilting of its head.

Fig. 5.—Coxa valga deformity is shown.

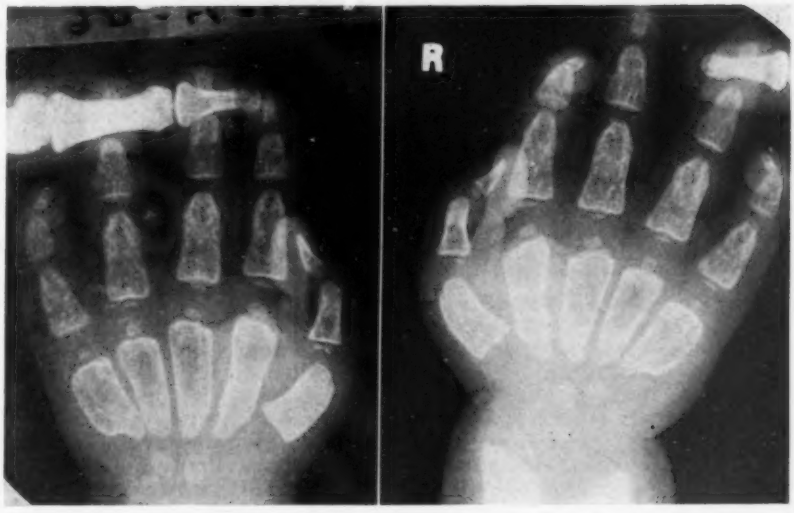


Fig. 6.—Note the characteristic deformities of metacarpals and phalanges; also shown is the tilting of the ends of radius and ulna toward one another.

deformities. It is his opinion that cerebrospinal fluid, escaping through a defect in the roof of the developing fourth ventricle, gives rise to the formation of "blebs" in the subcutis. If these blebs are not absorbed, the pressure resulting from their accumulation in various hollow parts of the embryonic surface (e.g., orbits, root of nose, blind sacs of limb buds) upsets the developing organs' balance and leads to deformities. A pituitary-hypothalamic derangement may also be due to similar increased pressure and so cause an upset in lipoid metabolism.

Whatever the etiology, the diagnosis in such typical cases as the one presented should not be difficult, but in less well defined instances other forms of retardation, dwarfism, and skeletal abnormalities must be considered, e.g., cretinism, rickets, syphilis, Morquio's disease, the lipoidoses. Excellent differential diagnoses are given elsewhere.[1, 5, 10-12]

SUMMARY

Lipochondrodystrophy is discussed and a case report in a negro child is included.

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# HERNIATED OVARY IN AN INFANT

NORMAN DUREN, M.D., F.A.C.S., AND S. ROSA FRANK, M.D.
BEAUMONT, TEXAS

HERNIATION of an ovary into the inguinal canal is an unusual finding. M. Ferro and Y. Ferro reported a similar case of "Congenital Inguinal Hernia of the Ovary," in 1910.* A review of the varying symptomatology is given in that paper.

## CASE REPORT

Baby C., a 4-week-old girl infant, was referred by her attending pediatrician, Dr. Frank. The infant's mother discovered a swelling in the left groin the day before admission to the hospital. The infant had appeared to be in pain. She cried frequently and took her formula poorly. Bowel movements had been somewhat loose and flecked with blood and mucus. There had been no vomiting.

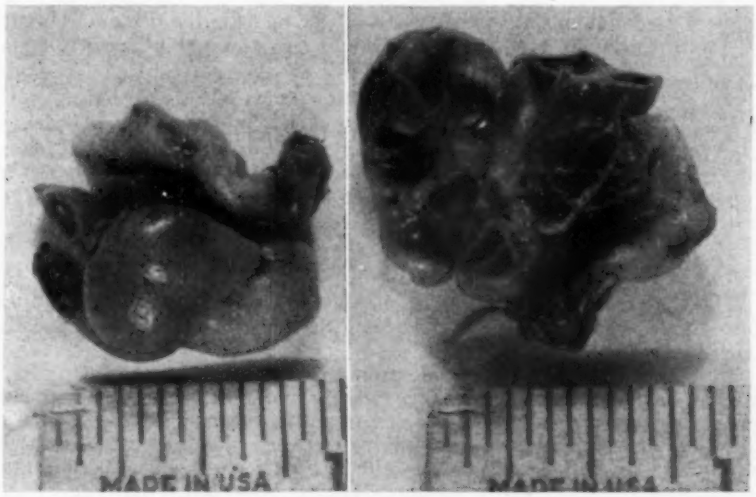


Fig. 1.—A, Ovary and overlying tube; B, ovary sectioned, showing cysts.

The infant did not appear acutely ill, although restlessness and discomfort were apparent. Abdominal distention was not marked. Digital examination of the rectum released gas and liquid stool. The tender mass in the left inguinal region was firm, movable, and about the size of a pecan. It was situated above Poupart's ligament near the upper end of the labial fold. It was irreducible. A scout film of the abdomen was negative. The blood count was normal. A preoperative diagnosis of herniated ovary was tentatively made.

Operative exposure under local anesthesia revealed a congested, edematous ovary and tube lying in hernial sac. The ovary and tube were ligated, excised, and the canal closed. Pathological examination of the specimen showed a congested cystic ovary. Postoperative recovery was rapid.

*Ferro Y., Ferro, M.: Congenital Inguinal Hernia of the Ovary, Med. de los ninos, 11: 17-20, 1910.

# The Social Aspects of Medicine

I wonder how many readers of the Column are aware that the Canadian Medical Association recommended compulsory health insurance to the Canadian House of Commons as far back as 1943. It is extraordinary that this important act on the part of organized medicine in our close neighbor should not have had the prominence which is its due in the medical periodicals, in particular in the *Journal of the American Medical Association,* since what organized medicine does in Canada should be of vital interest to organized medicine in the United States. The "Submission" by the Canadian Medical Association is too long to be given in the Column in its entirety. The parts which I have deleted are, however, the least important to us. Whatever is given is exactly as the text stands in the original.

What the Canadian Medical Association does is of great moment to pediatricians, for is not Canada a vast country of Anglo-Saxon heritage faced with essentially the same medical care problems as ourselves, and is not Organized Medicine in Canada composed of the same kind of doctors as organized medicine in this country? Why then should they think so differently from us?

E. A. P.

## A SUBMISSION RESPECTING HEALTH INSURANCE*

*Presented to the Special Committee on Social Security of the House of Commons*

By the Canadian Medical Association, April, 1943

*Mr. Chairman, Ladies and Gentlemen:*

The Canadian Medical Association welcomes this opportunity of appearing before the Special Committee on Social Security to discuss the problem of Health Insurance.

In respect to great national problems such as Health Insurance, The Canadian Medical Association has not been content in the other provinces to canvass its members only. On the contrary, the medical profession of Canada as a whole has been invited to make its voice heard in respect to these broad questions, to the end that it can be said without fear of contradiction, that the representations which The Canadian Medical Association now makes to the Parliamentary Select Committee express the considered views of the medical profession as a whole throughout the Dominion.

### Purpose of Submission

1. To emphasize that adequate medical care is essential to the welfare of Canada;
2. To review certain factors which handicap the medical profession in providing adequate medical care;
3. To point out that highly desirable, preventive and public health services are now inadequate;
4. To indicate the position of The Canadian Medical Association with respect to State Health Insurance;

From the Secretarial Offices of the Canadian Medical Association, 184 College Street, Toronto, Ont.

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5. To set forth features which should be included in any Plan of Health Insurance;

6. To point out the desirability of avoiding certain weaknesses observed in Plans of Health Insurance in other countries;

7. To make general comments with respect to a number of features to be considered in setting up any Plan of Health Insurance;

8. To show that Health Insurance can suffice to meet the medical needs of sparsely settled communities;

9. To discuss possible future developments in the years to come in the provision of good health care.

### I. ADEQUATE MEDICAL CARE ESSENTIAL

It is most essential to the welfare of our people that the medical care provided be not only of the highest standard but be readily available to all. Although our present system of providing medical care is characterized by many desirable features worthy of retention, it is recognized that there are a number of ways in which provision is not made for the full needs of the people at the present time.

To achieve its full destiny Canada must have healthy citizens. Our people can only enjoy full and vigorous health if there be provided for them adequate facilities for medical care (preventive and curative) of the highest standard, which will be readily available to all, irrespective of geographic location and financial status.

It would appear that certain changes are necessary, and to achieve this result the medical profession stands ready to cooperate, for the welfare of the people has always been its primary objective. At the same time the medical profession is proud of the achievements of those who have laid the foundations of medical practice over the centuries. The medical profession has served the people well—caring for the poor, placing patient before self whether it be in time of plague or battle, developing medicine from discovery to discovery, fostering research, preserving what is perhaps the highest code of ethics in the world and attracting to its ranks many of the keenest minds of each generation. It is in the interests of our people that these features of medical practice be preserved.

### II. HANDICAPS OR WEAKNESSES OF THE PRESENT SYSTEM

(a) The costs of sickness have become an increasing burden to many people, particularly those of moderate income, and have frequently prevented them from taking proper advantage of early diagnosis and treatment.

The extent and value and accuracy of medical knowledge and skill have increased tremendously, but at the same time the *cost* of this service has also become an increasing burden. The major increase has been due to the increasing complexity and delicacy of diagnostic methods and to the increased utilization of intricate apparatus and highly skilled personnel in treatment.

The diagnostic and "laboratory" equipment of Confederation days was limited to a stethoscope and (sometimes, not always) a thermometer. This equipment was the forerunner of the elaborate, complicated and costly, but highly accurate, equipment of our x-ray, pathological, biochemical and other laboratories. The number of hospital beds in Canada has increased from about 400 at the time of Confederation to over 100,000 at the present time. The investment in Canada is of some $250,000,000. In five or six decades the cost of providing hospital service which would be abreast of expanding medical knowledge has gone up from 75 cents per patient per day to an average of around $3.00 per patient per day. The cost of medical education has increased five or six fold and the time required has been more than doubled.

Medicine has ceased to be a science in which any one man can expect to be able to offer full service. For special diagnosis or for certain curative measures, individuals with special knowledge in selected fields must be called in. This has increased accuracy of diagnosis and efficiency of treatment but has added to the cost.

True, it could be pointed out that the results attained have more than compensated for additional costs. Better and earlier diagnosis and more effective treatment have shortened illnesses and saved lives beyond computation. Actually, despite increased costs of hospitalization, certain specific illnesses, such as pneumonia, gall bladder operations, etc., cost the patient less for hospitalization today than at the turn of the century, due to a greatly reduced period of hospitalization. Moreover, costs of medical care have not risen as rapidly over the years as has the general cost of living. Nevertheless, owing to the unpredictability of most illnesses and accidents and the reluctance or inability of most people to budget for illness, some means is necessary of relieving the individual of the burden of the cost of illness at the time when he can least afford to pay these costs.

The section of the community which has suffered most from the increase in cost is the great mass of honest thrifty folk of moderate means. The so-called "indigent" has usually received the medical care he has needed and illness has seldom created a grave financial problem for the well-to-do. But for the man of low or moderate income who desires to pay his way, the possible costs have definitely deterred him in many cases from seeking early advice or agreeing to the proper treatment.

*Voluntary medical and hospitalization plans.*—In an effort to alleviate the financial burden of sickness, a number of voluntary medical and/or hospitalization plans have been set up. The better hospitalization plans are known as "Blue Cross Plans." These voluntary contributory plans for medical care, or hospitalization, have been of much assistance to their members when overtaken by illness. Unfortunately there are many areas where these plans do not operate; also, being of a voluntary nature they do not cover all of the employed people of moderate means. Moreover, they seldom provide a complete service nor do they make provision for those who are unable to make any contribution.

(b) The distribution of medical services in the various parts of Canada, urban and rural, is not as it should be if all of our people are to receive full and adequate medical care.

This applies also to health services in general.

A major problem in medical distribution is that of providing adequate medical care in rural districts. Present wartime conditions are abnormal and, therefore, cannot be taken as a basis of discussion, but long before the war it was obvious that there was a heavy concentration of medical practitioners and of health services in general in the urban areas. Now with large numbers of rural doctors in the armed forces, the problem is much more acute.

This situation not only makes active treatment difficult to obtain, but it interferes with preventive services. For years infant mortality has been much higher in rural than in urban areas; the same applies to tuberculosis.

*Some Concentration in Cities Logical*

It should be pointed out, however, that, under any system, some concentration of medical personnel must be expected. Specialists could not be located other than in centres large enough to afford them an adequate clientèle; although located in centres, they serve both rural and urban patients. As the more technical or specialized diagnostic and curative procedures can best be done in hospitals, patients must continue to be taken to hospital for such purpose. Frequently the equipment or skilled personnel is only available in large urban

hospitals. As it is neither possible nor advisable to set up fully equipped and staffed hospitals in all rural communities, many rural patients will continue to be treated in the larger centres, irrespective of any plan which may be put into effect.

Moreover, it has long been noted that many patients in rural areas, even though a competent physician be available in the community, will drive beyond him to the city for diagnosis or treatment. Resulting from the advent of the motor car and good roads, many doctors formerly able to make a living in a country community have been forced to move to larger centres.

Board statements to the effect that a large number of rural municipalities are without medical care may convey an erroneous impression. A township or a municipality may not have a resident doctor, yet just beyond the borders of that area might be located several doctors serving the district in question with fully adequate care. Motor cars, improved highways and telephones have completely changed the former conception of what constitutes the geographic area for a normal rural practice.

(c) Many communities have failed to organize themselves in such a manner as to make the best use of existing legislation or to initiate their own health services.

Many communities have failed for various reasons to organize themselves to take full advantage of existing legislation which would permit them to develop much needed health services. For instance, in rural areas with inadequate voluntary non-profit hospitals, more municipal or "union" hospitals would have greatly improved health services. For various reasons, among which might be cited economic conditions, lack of understanding and leadership, and even indifference, the public health services in many areas have often been but nominal, or even entirely absent.

### III. PREVENTIVE SERVICES INADEQUATE

The present program of preventive medicine in the country is far from adequate. Our major emphasis in the past has been on the cure of disease—on *negative* health, as it were. There should be more emphasis in the future on *positive* health, on preventive medicine and public health. It is less costly to prevent disease than cure it, yet our progress in this direction, although steady and gratifying, has been far too slow.

In making these statements there is no implication in the slightest that the provincial, federal and municipal departments of health have not done excellent work. Actually they have accomplished much, frequently under considerable handicap. But the results have been much less than could have been achieved had adequate funds been available. It is unfortunate that, while, in the past, money has been freely available for so many other purposes, yet so much difficulty has been encountered in obtaining adequate funds for effective programs of preventive work and public education on health matters.

## Health Insurance

### IV. THE POSITION OF THE CANADIAN MEDICAL ASSOCIATION WITH RESPECT TO HEALTH INSURANCE

Realizing the situation, as already set forth, and that solutions for these difficulties must be found, The Canadian Medical Association has been actively studying the subject of Health Insurance for nearly fifteen years. In 1934 an outline of a possible plan of Health Insurance was drawn up and adopted by the Council of the Association. Studies continued and the principles upon which this outline was based have been reviewed and altered, or re-affirmed, from time to time up to and including the annual meeting of The Association in June, 1942.

*Principle of Health Insurance Approved*

At a largely attended special meeting of the Council of The Canadian Medical Association held in Ottawa, January 18th and 19th, 1943, the subject of Health Insurance was again considered. Previously the Council had said that if Health Insurance were to be established in Canada it should be along certain lines, but had refrained from recommending it or opposing it. At this time, and for the first time, the Council went farther. In Ottawa in January, 1943, the Council endorsed the principle by passing the following resolution:

WHEREAS the objects of The Canadian Medical Association are:

1. The promotion of health and the prevention of disease;
2. The improvement of health services;
3. The performance of such other lawful things as are incidental or conducive to the welfare of the public; and

WHEREAS The Canadian Medical Association is keenly conscious of the desirability of providing adequate health services to all the people of Canada; and

WHEREAS The Canadian Medical Association has for many years been studying plans for the securing of such health services;

THEREFORE BE IT RESOLVED THAT

1. The Canadian Medical Association approves the adoption of the principle of Health Insurance.
2. The Canadian Medical Association favours a plan of Health Insurance which will secure the development and provision of the highest standard of health services, preventive and curative, if such plan be fair both to the insured and to all those rendering the services.

V. FEATURES WHICH SHOULD BE INCLUDED IN ANY PLAN OF HEALTH INSURANCE

We visualize for Canada a system of Health Insurance which will be more all-inclusive, efficient and sound than any which has ever been devised and operated anywhere. It should place much emphasis on the prevention of disease and the development of a high degree of physical fitness, and should also include complete modern diagnostic and curative services. Possibly this full program cannot be immediately instituted in its entirety, because of shortage of trained personnel and of institutions, and possibly because of cost, but the full service should be visualized and planned. Medical knowledge in the prevention and cure of disease is far ahead of the means for its general utilization by the public.

It is obvious, too, that any plan of Health Insurance which is not supplemented by a program to ensure better nutrition, better housing and the reduction of worry and anxiety, particularly for those of low and uncertain income, will fail of its objective.

*A. Adequate Preventive and Public Health Provision*

1. Reorganization of public health and preventive services is necessary to *place more responsibility for various procedures on the family practitioners.* Their services must be available for and integrated with the public health officials' program. The family must be the unit and the family doctor the first line of defence in this program;

2. Our rapidly expanding knowledge of *nutrition* should be translated into action—adequate diets should be available to all. This implies not only a vigorous educational program but also a degree of economic security to which reference has previously been made

3. The present knowledge of the value of *inoculations, vaccinations and other immunizing and diagnostic procedures* in the control and eradication of

disease should be fully utilized by the extension of public health services, operating with the assistance of the General Practitioner;

4. General programs of *hygiene* and *sanitation* must be promulgated and carried out;

5. *Periodic examinations* at suitable intervals should be available to all. The early recognition of disease is the greatest weapon in its mastery;

6. *Complete examinations of all children,* should be linked with the means for correcting any deficiencies. This would greatly improve the health of our children;

7. A more general application of our present knowledge respecting *Goitre Control* could bring about a definite reduction in the frequency of this disease in affected areas. All such areas in Canada could be determined by careful study;

8. A well organized program to control *venereal disease* is necessary throughout the country.

9. Fifty thousand Canadians are now suffering from *cancer;* more than 12,000 die of cancer annually, yet many, if diagnosed and treated early, can be cured. An aggressive program to combat cancer should be an integral part of health insurance.

10. *Tuberculosis* might be eradicated from Canada in 25 years if facilities be provided for its early recognition and efficient control.

11. *Maternal Welfare.*—Maternal deaths and disabilities have been markedly lessened, but could be reduced still further by a maternal welfare plan which would incorporate all those features which have proven to be of value;

12. *Full dental care for children* is a prerequisite to good health;

13. *Pre-employment examinations* would help to direct individuals into employment which would be most suitable for them.

A progressive program for the improvement of the health and *physical development* of children and young people by supervised playgrounds and controlled exercises and physical fitness programs is desirable.

Also, the treatment of certain conditions is closely linked with the *housing* problem, and some integration of authority should be planned in this respect.

Such a general program would greatly assist in the fight to eliminate certain diseases such as diphtheria, whooping cough, tuberculosis, rickets, typhoid fever and smallpox, and would assist in the reduction in the frequency of many others, such as certain forms of mental disease, rheumatic heart disease, goitre, venereal disease and cancer.

As years pass the cost of curative services might be expected to materially decrease as disease is controlled and physical fitness increased.

*B. General Practitioner Services*

The fundamental service should be general practitioner service. When medical services are needed, such should be available without cost to the patient at the time. The individual should have the right to choose his medical adviser —and vice versa.

*C. Specialists and Consultants*

The science of medicine has progressed so rapidly that many conditions call for investigation and treatment by adequately trained and recognized specialists and consultants. This service is invaluable and must be freely available.

*D. Additional Diagnostic Services*

There should be available for all the people whatever diagnostic aid would be of value. This should include laboratory, radiological and other scientifically recognized diagnostic procedures and should also include consultant services. If

necessary, diagnostic facilities should be set up to serve designated areas. In some cases the pooling or centralizing of some of these diagnostic services could reduce overhead and consequent cost.

All necessary diagnostic aids which have been proven to be of scientific value should be available for all the people.

*E. Hospitalization*

Hospitalization accommodation should be available to all who need it and for such time as they need it.

*F. Nursing*

A visiting nurse service should be available in the home on the order of the medical adviser. Full time nursing service should be made available upon the authorization of the regional medical adviser.

*G. Drugs and Appliances*

Drugs and pharmaceutical preparations, as authorized in an official formulary which could be prepared, should be available upon the order of the medical adviser. Appliances, such as spectacles, crutches, artificial legs, etc., should be available within reason when authorized.

*H. Dental Services*

Dental prophylaxis and care with a stipulated limit should be available to all.

*I. Problems Needing Special Attention*

There is considerable need for the development of adequate facilities for the rehabilitation of those partially incapacitated by physical and mental illness. Such a program would be both humanitarian and economically sound.

There is need, too, in every province in Canada for more homes for the care of the aged and the infirm.

This also applies to the care of those who are chronically ill.

## VI. WEAKNESSES OBSERVED IN PLANS OF HEALTH INSURANCE ELSEWHERE SHOULD BE AVOIDED

In the setting up of any Health Insurance measure in Canada, it is highly desirable that we profit by the experiences of other countries. Their Health Insurance enactments have been studied carefully by our Association. To obtain first hand information, The Canadian Medical Association sent its General Secretary to Europe in 1937 to study Health Insurance in operation.

The following weaknesses and omissions observed there and elsewhere should be avoided in Canada:

*(a) Limited range of service in some countries*

A plan which provides for general practitioner service only does not provide a complete service. Such arrangement is not fair either to the patient or to those who must render specialist service.

By not providing for hospitalization, patients are either unable to obtain needed hospitalization, or must bear heavy financial burdens, or must accept charity. All too frequently an insurance scheme thus becomes a charity scheme.

*(b) Dependents not covered in some plans*

To omit from benefits the dependents of the insured, still requires the breadwinner to finance the major portion of sickness in the average family (somewhat over two-thirds of the total hospitalized illness in a family with two children and a still higher proportion—nearly four-fifths—in the case of home and office practice).

*(c) "Indigents" not covered in some countries*

The inclusion only of those who are employed, or who receive up to a certain income, still makes no provision for the varying but usually large number who are in the group frequently referred to as "indigent" or "near-indigent." From the viewpoint of national health, and particularly from that of preventive medicine, it is most important that this group be covered.

*(d) Friendly Societies as "Carriers" found undesirable*

The utilization of Friendly or Benevolent Societies as carriers under the Act in one large national plan has had unfortunate results. Although politically expedient at the time to obtain support for the measure, it has now become obvious that these carriers have introduced a third party between patient and physician and between citizen and government. Over the years these carriers have developed large surpluses and have now become so powerful that, through their membership, they can dictate to the government in insurance matters. In another country they have played politics and have been more concerned with the competition for members than with the medical services rendered.

*(e) Inadequate payment for services rendered*

Inadequate payment for services rendered can be of serious concern to the general public for eventually it will result in a lowering of the quality of the service rendered. Physicians, dentists, nurses, technicians and all those rendering specialized or expert service should be compensated in accordance with their investment in years and in money in initial and post graduate training, and with their responsibilities and the arduous nature and dangers of their work. It is vitally important to the health of the nation to ensure that medicine of the future shall continue to attract a large percentage of the keenest young men and women of each generation.

*(f) Inadequate provision for necessary diagnostic services*

Some plans do not make provision for adequate diagnosis, such as laboratory and x-ray examinations and consultant services. When required these must be paid for by the patient or, as is frequently the case, be provided on a charity or part-pay basis by hospitals and consultants. This is not conducive to early diagnosis, effective treatment and prompt recovery.

*(g) Lack of democratic principles in some plans*

In one well established plan in a major European country the methods adopted are exceedingly dictatorial and bureaucratic and permit the individual member or those rendering the service little, if any, opportunity for stating grievances or correcting omissions or abuses. Health Insurance should be a well integrated partnership, with all parties concerned—the insured, those rendering the services and the state—given a voice in its direction.

*(h) Preventive medicine and Public Health not emphasized in other plans*

Of utmost importance, most existing plans of Health Insurance do not lay sufficient stress upon preventive medicine. The emphasis has been largely upon curative medicine. Canada has an opportunity to develop a system of Health Insurance which, by the inclusion of the preventive features already mentioned, can not only set new standards of Health Legislation, but can have an effect on our future national health far beyond our comprehension.

## VII. ADDITIONAL FEATURES TO BE CONSIDERED IN ANY PLAN OF HEALTH INSURANCE

(a) In developing a Health Insurance plan in Canada great care should be taken to ensure that the proposals are sound and sufficiently comprehensive.

The medical profession recommends a complete service of high standard. To the medical profession it will mean great and in some respects unwelcome changes. We are, however, willing to assist in its formation and its operation, *provided* that it will secure for all the insured a high standard of service, as high

or higher than has been available in the past, and will be fair both to the person insured and to those rendering the services.

(b) Should Health Insurance be a strictly federal measure or should it be a provincial measure, operating under federal enabling legislation and with federal assistance?

We realize that there are advantages in a single nation wide measure of Health Insurance. It does appear, however, that there are certain factors which would indicate that Health Insurance should be introduced on a provincial basis. Provincial plans should be co-ordinated by federal legislation of an enabling nature. Such federal legislation should outline those standards to which provincial enactments would be expected to conform in order to receive federal subsidies. Our reasons are as follows:

1. Local conditions, such as costs of living, needs for and costs of services, incomes, etc., vary in different provinces.
2. Some degree of variation and experimentation in detail is not without value.
3. It would be much more difficult to draft a federal measure acceptable in detail to all parts of Canada, than to prepare a federal measure which would outline the general provisions only and leave many of the details to be determined in the individual province.
4. By maintaining uniformity in the general structure of the plans to be developed in the different provinces, the problems of later unification, should that prove desirable, would be simplified.
5. Revision of the British North America Act would not be necessary.

*Some Form of Federal Control Essential for Uniformity*

If Provincial autonomy is to be maintained as suggested above, it is extremely important that a sufficient degree of federal control be retained. This control should be limited to such matters as the extent and the standards of the services, as indicated previously. It is important, however, that control be maintained as it is only through this means that a satisfactory degree of uniformity can be obtained. Unless there is considerable similarity in methods and program in matters of disease prevention and control, the results from the national viewpoint will be jeopardized.

(c) Administration.

*Federal.* If it eventuate that the federal legislation passed be of an enabling nature, it would follow that the function of the federal government with respect to provincial enactments would be broadly to supervise the operations of these provincial acts in order to determine the eligibility of the provinces to receive the federal subsidies and other assistance provided under the Act.

On this basis it is recommended that the federal administration be under the Minister of National Health, with an Advisory Dominion Council on Health Insurance.

If federal administration is to be carried out within the Department of Pensions and National Health it should be under a Health Insurance Division, with a Director in charge.

*Provincial.* In the provinces it is strongly recommended that the plan come under a *non-political independent commission* which might be responsible to the legislature through the Provincial Minister of Health. Because of the vital nature of health care The Canadian Medical Association considers it highly desirable to remove these important health services from the possibility of interference in the continuity of services or personnel by the vicissitudes of political change.

We believe, moreover, that the Commission should be so constituted, appointed and empowered that it will possess ample freedom of action and that its

members will be representative of and preferably nominated by the various professional and other groups interested in the operation of the Act.

(d) Certain details might wisely be left to the Province.

Among these might be:

1. The income level (which might be high enough to include all the residents, or set at an agreed level which might vary in the various Provinces) below which the residents would be included under an obligatory plan;

2. The rate and method of payment for all services, professional and institutional;

3. The best means of obtaining full preventive and public health services within the Province concerned;

4. The particular means which might be necessary to safeguard the basic necessity of maintaining adequate teaching services in connection with Medical Faculties and Teaching Hospitals within the Province.

(e) The patient of little or no income should be included.

Those patients, frequently termed "indigents," who cannot pay for adequate health services should be included. It would be grossly unfair, not only to the public, but also to those rendering the health services, to omit this group from the benefits of the plan. Provision for the inclusion of such patients should be specifically made in the federal enabling and provincial Acts.

(f) The Plan should be on a contributory basis.

The principle of having those participating in the benefits contribute to the Fund would seem to be sound. The individual who shares in the cost of providing the benefits is much more likely to co-operate in keeping down unnecessary calls upon the Fund. For those not employed by others, a basis of direct assessment could be worked out.

(g) Remuneration.

The remuneration of those rendering the services required under this Plan should be reasonable and in conformity with the high standards of service expected of them. In the case of the medical practitioner (general practitioner), it is obvious that some variation in the basis of remuneration may be necessary in different areas. In some areas a "fee-for-service" basis may be desirable; in others a "capitation" basis (so much per annum per individual on a panel, irrespective of the amount of service rendered) may be preferable; in other areas, particularly certain rural areas where the population is small and scattered, a "salary" basis, or a combination of any two of these three alternative methods may prove desirable. Because of this situation it is recommended that the method of remuneration, as well as the amount, be left for decision in the individual province after consultation between the Commission and representatives of the medical profession.

In the case of the specialist or consultant it would seem necessary to follow a fee-for-service basis.

(h) Medical Education must be maintained at a high standard.

Fully trained medical personnel in sufficient numbers are absolutely necessary to any plan of general Health Insurance. Such are necessary for the health welfare and the future of the nation. If anything happens to the proper teaching of succeeding generations of doctors, scientific medicine dies and with it declines the health of the nation. The truth of this assertion is obvious.

Heretofore clinical teaching has been carried on in wards set aside for that purpose; these have usually been the public wards of our large teaching hospitals. The hospital service has either been free to the patient or at minimal rates and the medical attention has been without charge. Although the patients have been available for the bedside discussion and demonstration of methods of examination and treatment, the service has been of the highest standard of excellence and always with the complete co-operation of the patients concerned. Under a Health

Insurance plan in which there would be no longer any group of individuals who would receive so-called "free treatment," there is a possibility that opportunities for clinical teaching may be reduced. While it is anticipated that the long recognized excellence of the quality of diagnosis and treatment in teaching hospitals may alone suffice to ensure a sufficient number of patients, it must be recognized that it is distinctly in the public interest that the popularity of teaching hospitals should be maintained, to the end that teaching may not be curtailed.

(i) The Plan should be on an obligatory basis for the income groups specified and should be without exceptions.

In other words all persons coming within the income brackets stipulated should be required to contribute to the Fund and be eligible for benefits and there should be no provision for the exclusion of groups below the income level set for the operation of the Act.

Much of the value of a fully adequate scheme lies in its *preventive* services. The entire population benefits from the functioning of these services and should contribute to them. Conversely, if preventive measures are not applicable to any groups in a community, not only do these groups suffer, but they may constitute a health menace to others.

There are certain plans operating in parts of Canada with voluntary prepayment for health services, providing either medical care or hospital care, or both. Some of these plans are excellent, as far as they go. But none of these plans, to the best of our knowledge, offer the complete protection, preventive, diagnostic and curative, which we believe in envisioned in this proposal. Some of these plans do not cover dependents. Others cover dependents but, in one-industry towns, may not cover the scattered townspeopls not directly connected with the plant. Any acceptable national or provincial plan should offer more than can any individual plan. These plans deserve great credit for their pioneering service but it is in the national interest to have all single plans amalgamated. With a government subsidy, the resultant cost to these industries should be less than under present conditions.

It has been the experience in other countries, notably Great Britain, that the exclusion of certain groups of individuals and the utilization of existing plans has complicated the situation and made it more difficult to attain ideal results. From the viewpoint of administration, a multiplicity of local arrangements, perhaps overlapping in the one community creates a situation so chaotic that efficient operation becomes very difficult [see page 18, par. (d)]. Another difficulty arises when an individual changes his employment or moves to another town. If Canada is to have Health Insurance, let its action be not hamstrung by lack of vision or courage.

There are within Canada, also certain groups who do not accept medical services—at least at times. They all, however, have been the beneficiaries of preventive and public health services and most of them, sooner or later, of diagnostic and treatment procedures as well. Apart from this, however, all our citizens have a *common community obligation*. There is probably no argument which can be advanced in support of the exclusion of certain groups from participation in any Plan which was not advanced two or three generations ago in opposition to the inclusion of the general population in a scheme under which all were taxed to support our great Public School system, even though all do not avail themselves equally of its privileges. It should be quite proper, of course, for certain individuals, if they so desire, to obtain their health care outside of the provisions of any insurance scheme, even as individuals now use schools which are apart from those provided under the authority of our Public or Separate School Acts.

It should also be proper for any institution or any member of any profession to decline to act under the Plan and to offer services in a private capacity under any arragnements which are agreeable to themselves and to their patients provided these arrangements are not contrary to public interest.

(j) If cash benefits be provided for loss of time through illness, such should be through a separate fund entirely apart from the Health Insurance Fund.

Cash benefits, obviously, are a matter of great importance to the insured. It is entirely true "that a person sick needs financial assistance more than a person well." This is realized, yet from the viewpoint of conscientious medical service cash benefits incorporated in Health Insurance have introduced serious complications.

In every country where cash compensation for loss of time has been a part of a sickness insurance plan, there has been great dissatisfaction over the complications of operation which have developed and the drain on the common fund because of its inclusion. Because of experience gained in other countries operating a Health Insurance plan, we strongly recommend that such assistance be provided through some plan to be devised *entirely apart* from the Health Insurance plan.

### VIII. CAN HEALTH INSURANCE MEET THE MEDICAL NEEDS OF SPARSELY SETTLED COMMUNITIES

The answer is "yes." In areas where a doctor could not make an adequate living on the ordinary basis of remuneration under the plan, the Commission could provide a salary adequate enough to prove attractive to our doctors. If district nursing service be provided, as well as adequate diagnostic and reasonable hospitalization facilities, and if reasonable opportunities to get away for post graduate study or for vacation be made available, our doctors will go to rural areas. If bursaries be made available to permit brilliant young people without adequate financial means to complete the medical course, their services in such areas for a definite period of years could be required. The same effect could be obtained, as has been done in Australia, by offering young men and women opportunities for post graduate study upon the completion of a definite period of service in rural communities.

### IX. HEALTH AND HEALTH PRESERVATION IN THE YEARS TO COME

During the past two generations more progress has been achieved in making good health care available to all than for the two previous centuries. We anticipate still greater progress in the years to come.

#### *Improved Facilities for Diagnosis*

Better and more readily available diagnostic procedures will do much to still further reduce illness. It is hoped that the present commendable system in many provinces of having certain swabs, smears, blood samples, etc., examined by the Department of Health will be extended. The setting up of branch laboratories has been of tremendous assistance. The establishment of "diagnostic centres" in strategic locations may be anticipated. These could be governed by adequate regulations relating to the nature and quality of service, charges and remuneration and to ethical relations. As the inclusion of people of inadequate means under a health insurance plan may make most hospital out-door departments unnecessary, some of these facilities might be converted into "diagnostic clinics" or centres for referred patients.

#### *Rural Medicine*

Rural medicine will be better organized in years to come. Good care must be available to all. This does not mean that a highly trained doctor must sit

around waiting for a handful of scattered settlers to call for his services, but it does mean that methods will be worked out whereby "positive" health care can be made available to these people. This can be accomplished by adequate district nursing, by a reasonable system of rural hospitals and by providing doctors with sufficient returns to compensate them for the hardships and strain of that type of practice. This will all be made easier by providing good roads and improved methods of transportation.

As for *specialist* and *consultant services,* there is no reason why such cannot be made available to rural areas. To a large degree the removal of the economic barrier and better transportation will permit readier consultation at the doctor's office. The bigger problem—that of bringing the specialist or consultant to the rural patient—could, and may be, overcome in a fashion similar to that evolved some years ago in Australia and now widely utilized in military medicine, that is, by *aerial transport.* The day may not be far distant when health service planes could be made available in selected areas to carry specialists and consultants, or even "surgical teams," to seriously sick or injured patients. The same planes could function as "aerial ambulances" to carry patients to hospital. The helicopter type of plane would be particularly valuable for this purpose.

*Health Centres*

In certain areas, we can foresee the creation of "Health Centres" such as have been envisioned by the British Medical Planning Commission in its constructive report in 1942. In essence the proposal is as follows:

The Health Centre would be a building providing a series of consulting rooms, waiting rooms, a small operating room for minor surgery, laboratories and other accommodation. The general practitioners in the town, or that part of a city, would use these facilities for their office consultations. They would not be in partnership, as in a clinic, but would merely use these facilities in a co-operative manner. Each doctor would have his own patients who in turn have exercised their right to select their own medical adviser. Specialists would see patients either in the centre or elsewhere. Serious or more difficult cases would be sent to the hospital. Work of the centre would be preventive and educational, as well as diagnostic and curative.

The x-ray department and the laboratories would be available to all participating doctors: so would the dispensary, the secretarial department and the clinical records department. Health visitors and district nurses would have their headquarters at the centre.

A large city might have a number of these centres. We might add that where a hospital is centrally located it would be quite possible to have such office building adjacent to or part of the hospital itself.

*Hospitals*

Our present hospital system is excellent, but it will probably show better integration in the years to come. We sincerely hope that we shall always continue to benefit from the services of the great group of voluntary hospitals, lay and religious, which have served our sick for many years. It should be possible, however, without jeopardizing this system, to have hospital construction and equipment in general follow a carefully planned program based upon provincial and community needs rather than upon unrelated and sometimes overlapping local efforts.

Specially designed hospitals should be available in rural areas. These should not be made more elaborate than is justified by local needs, by professional and technical staff available and by the proximity or otherwise of similar or better facilities. Nor should they be more numerous than is necessary to meet local

conditions. As transportation facilities improve the need for numerous rural hospitals should decrease rather than increase. Moreover, if rural and urban hospitals could be so linked that patients could be quickly transferred when necessary to institutions with more elaborate equipment, the net result should be a reduction of mortality, more rapid convalescence and a definite saving of both skilled personnel and special equipment.

One regrettable gap in our present facilities will surely be corrected—more hospitals for convalescent patients and for those suffering from chronic or incurable illnesses. Actually such provision would be a distinct economy.

Should these improvements be combined with general increase in social security and health planning—assured employment and maintenance, better nutrition, better sanitation, better housing and planned physical programs—the improvement in national health would be amazing.

## Conclusion

Throughout the last ten years, The Canadian Medical Association has consistently and repeatedly sought to ascertain the opinion of the medical profession on the subject of Health Insurance. The views expressed in this submission crystallize those opinions.

Since this memorandum was prepared, there has been placed in your hands the Draft proposals of the Advisory Committee on Health Insurance. While some of us have had the opportunity of seeing these proposals, they have not been seen by our constituent societies. It is our hope that in the immediate future the medical profession throughout Canada will be permitted to examine these proposals in detail and that shortly thereafter we may be in a position to come back to you to discuss those aspects of the proposals upon which you might desire our advice and opinion.

In conclusion, The Canadian Medical Association desires to assure the Special Committee on Social Security that our entire organization stretching from sea to sea stands ready to render any assistance in its power towards the solution of one of the country's most important problems, namely, the safeguarding of the health of our people.

# Report of Meeting

## THE FIFTH INTERNATIONAL CONGRESS OF PEDIATRICS

Despite four hot and humid July days in New York City, the meeting of the Fifth International Congress of Pediatrics at the Waldorf-Astoria Hotel was most successful. The total registration reached over 2,200, against an estimated expectation of 1,500, and more than 400 representatives from over sixty foreign countries were present. From every viewpoint it was an outstanding success. The complete transactions will be published at some later date in the *Acta Pediatrica*, and hence our report is merely reportorial in character.

The most outstanding feature was the scientific exhibit, which spread beyond the ballroom floor of the Waldorf to distant parts of the hotel. Approximately 150 exhibits of the work and developments in our pediatric clinics were shown, and it would have required many days to give them the study they deserved. Obviously, months of work had been given to the perfection of the exhibits, whose graphic picture of the extent to which pediatric research has reached in this country in the past ten to fifteen years was overwhelming. Without comparing or in any way reflecting upon those unmentioned, it may be said that certain exhibits or groups were of particular interest. In the Wedgewood Room, far from the meetings, was a splendid exhibit from a number of clinics of the newer work on cardiac and intrathoracic surgery. Another was the exhibit on endocrine disorders of childhood and adolescence which a number of workers in other cities had joined with Dr. Lawson Wilkins of Baltimore in producing. The exhibit of the Virus Research Laboratories of the Children's Hospital of Philadelphia was most interesting, not only for the work but for the technical, graphic way in which it was presented. After the first day, many of those in attendance spent their time in the exhibits rather than the scheduled meetings. It was rumored, and we hope the rumor is true, that some thirty or more of the exhibits are to be microfilmed and thus made available for the more careful study they deserve and for teaching purposes in our medical schools. It would be a most valuable contribution and a worth-while, permanent memorial of the congress. To the many pediatricians who spent hours arranging the material for the exhibit and for the energy and work of the committee in charge, our sincere congratulations. Had there been no other program, the exhibits alone would have made the congress worth attending.

At the formal opening of the congress on Tuesday morning, July 15, addresses of welcome were made by Dr. Henry Helmholz, President of the Congress, Commissioner Bernecker, representing the Mayor of New York, Dr. Thomas Parran, Surgeon General, United States Public Health Service, and Miss Tenroot of the Children's Bureau. Dr. Parran read a message to the Congress from President Truman, which was as follows:

"The United States is honored to be host to the Fifth International Congress of Pediatrics. We welcome particularly the distinguished scientists from other lands who have come here to pool their knowledge with that of American pediatricians so that children everywhere may lead healthier lives. To promote the health of children the world over is a part of our task of building permanent world peace."

General Frederick H. Osborn, Deputy Representative, United Nations Atomic Energy Commission, then welcomed the delegates on behalf of the United Nations, and, in a brief but forceful talk, contrasted the work of his commission in its efforts to control the destructive forces of atomic energy with the work of the pediatricians of the world gathered to improve the welfare of children and the future generations of the world. He stressed the blocking of world unity by a certain ruling group of 10 per cent of the world population, and expressed the thought that, as, after the religious wars of past centuries, man had learned that peoples with religious differences could live side-by-side in peace, so man must learn that peoples with different ideological economic theories can live in peace, and that war for economic and social ideologies is unnecessary.

The first plenary session, on the subject of nutrition, was held on Tuesday afternoon. Professor R. A. McCance of Cambridge, England, discussed the findings of the British Research Council Unit, which studied the nutritional condition of children in the British Zone in Germany. Despite the low caloric intake provided for children, he did not feel that any serious damage had been done and that the nutritional health of German children was not a matter for present concern. Dr. J. P. H. Jonxis of Rotterdam, Holland, discussed the nutritional status of the Dutch children and the studies which had been carried out on their nutritional needs. He concluded that the lack of Vitamins A and D and of animal proteins were the serious deficits in the diet of children in Holland as a result of the war, and that these were of greater importance than the question of calories. Dr. J. Gillman, of Johannesburg, South Africa, discussed and presented interesting slides of childhood pellagra. Dr. P. Plum of Copenhagen, Denmark, discussed celiac disease, the incidence of which increased markedly in Norway, Denmark, and Finland during the war, but not in Sweden, which was an unoccupied country. This led to the impression that some nutritional factor was of etiologic importance in the disease. Dr. G. Logaras of Athens, Greece, discussed the nutrition of children in Greece and the curtailed food supply. Dr. D. Riis of Oslo, Norway, described conditions in his country. Throughout the war, due to planned measures of food allotment, the babies in Norway remained in good condition, but the nutrition of children of school age was impaired with a marked decrease in height and weight growth compared with prewar standards. An interesting observation was a decrease in dental caries in the school-age children. This observation is in agreement with the findings in a study of Italian children which will appear shortly in the JOURNAL.

The discussion at the plenary session on Wednesday morning was on three subjects. The first subject was "Diseases Caused by Filtrable Viruses." Dr. A. Sabin of Cincinnati, Ohio, summarized our knowledge of neurotropic viruses, with emphasis on poliomyelitis, and Dr. Thomas Francis gave the principal talk on the respiratory viruses. The second subject was "Chemotherapy of Infectious Diseases." The principal speaker was Dr. Chester S. Keefer of Boston, who summarized the uses of streptomycin. Professor G. Frontali of Rome, Italy, discussed penicillin, and Professor R. Degkwitz of Hamburg, Germany, described what he termed "aimed injections" of drugs into such organs as the liver, spleen, and bone marrow. Such drugs must be insoluble in water and crystallizable.

The third topic was "Neonatal Mortality," with Professor Alan Moncrieff of London, England, as the principal speaker. He discussed the diminishing birth rate in England and the necessity of reducing the infant mortality rates, which led to a study of the causes and the instigation of measures to reduce

infant mortality in that country. Dr. C. Gyllenswärd of Upsala, Sweden, considered the problem of neonatal mortality one of medicine as well as social medicine. Professor A. Reuss of Vienna, Austria, discussed the medical causes of deaths in the neonatal period.

On Wednesday afternoon, sectional meetings were held in the building of the New York Academy of Medicine, which has just finished celebrating its Centennial. Sectional groups were held for the discussion of:

I. Factors in pregnancy affecting the child
II. Preventive pediatrics
III. Bio-immunological procedures
IV. Vitamin requirements and avitaminosis
V. Insect and other vectors of disease
VI. Rheumatic fever
VII. Endocrinology in childhood
VIII. Miscellaneous topics

The plenary session on Thursday morning, July 17, was given to a discussion of tuberculosis. Chief interest centered on the use of BCG. Dr. J. Wallgren of Stockholm, Sweden, feels that vaccination with BCG fills a gap in the fight against tuberculosis and compliments our present preventive methods for tuberculosis. Professor H. Jonscher of Poland thinks BCG should be used where there has been a nearly complete breakdown of public health and hospital facilities, as in Poland. Dr. A. Chattás of Cordova, Argentina, was most urgent for its use on a large scale.

The final plenary session on Thursday afternoon discussed incompatibility of blood, with Professor G. De Toni of Genoa, Italy, as the relator, and alimentary toxicosis as a second subject, with Professor E. Kerpel-Fronius of Pécs, Hungary, as relator.

On the social side, there were numerous activities. Among those were daily trips to sessions of the United Nations. On Wednesday afternoon some 1,500 of those present had a boat ride on the Hudson River as far as West Point. Supper was served on board, with dancing on the way back. It was a happy but tired crowd that piled into busses for the hotels at 11 P.M. The congress ended with a banquet at the Waldorf on Thursday night, at which the Findlay medal, a Cuban award, was presented to the President of the Congress, Dr. Helmholz. Among the speakers at the banquet were Dr. Lee Forrest Hill, president of the American Academy of Pediatrics, Dr. Wallgren, Dr. Escardo of Montevideo, Uruguay, and Dr. Robert Debré of Paris.

Nearly 100 of the foreign delegates are making a post congress tour of the midwest, visiting Cincinnati, Chicago, Rochester, Minn., Detroit, and Rochester, N. Y., while a smaller group is visiting clinics and points of interest in the East.

The planning and arrangement of the details of the congress were the work of many individuals. We dislike to single out any one person, as all shared in making it a success, but we feel that particular recognition should be given to Dr. L. Emmett Holt, Jr., of New York, the general secretary of the congress, who, for many months, has given his time and energy to its planning; that it took place was due in no small part to his work.

There is one other factor which should be widely known. Due to economic conditions prevailing in many countries, and currency and exchange restrictions, it was necessary to help meet the traveling expenses of a number of foreign delegates. A sizeable sum of money was raised for this purpose aside from the actual expenses of the meeting. The pediatricians are deeply in-

debted to those who contributed so generously to this fund; hence we close this account of the congress with thanks to the following:

American Friends Service Committee
American Red Cross
American Relief for Czechoslovakia
American Scandinavian Foundation
English-Speaking Union
Otto Grob
Help Finland, Inc.
Institute of International Education
Kosciuszko Foundation
Lessing Rosenwald Foundation
Josiah Macy, Jr., Foundation
Masaryk Institute
Samuel R. Milbank
Milbank Memorial Fund
New York Foundation
Oberlander Trust
Frederick Osborn
Pabst Foundation
Alexander Pathy
Rockefeller Foundation
Save the Children Foundation
Judith Schoellkopf
George Skouras
Stephen Stephanides
Uihlein Foundation
United Yugoslav Relief Fund
Unitarian Service Committee
World Health Organization Interim Commission

A number of business firms consented to become patrons of the Congress by contributing to its cost.

These firms are:

The Gordon Armstrong Company
Beachnut Packing Company
Burroughs, Wellcome & Company
Johnson & Johnson
Lederle Laboratories
M & R Dietetic Laboratories
Mead Johnson & Company
National Dairy Products Company
Nestlé's Milk Products, Inc.
Procter and Gamble Company
Swift & Company
White Laboratories

# News and Notes

The **American Board of Pediatrics** will hold fall examinations on Dec. 5, 6, and 7, 1947, at Dallas, Texas, and on Jan. 9, 10, and 11, 1948, at Philadelphia, Pa. For further information write The Secretary, 718 Royal Union Bldg., Des Moines, Iowa.

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The Public Health Report of June 20 summarizes the incidence of poliomyelitis in 1946 when 25,191 cases were reported. This is a greater number than for any prior year except 1916, and an increase of 85 per cent over 1945. The disease was of epidemic proportion in the North Central States and was high in some of the neighboring states. Minnesota had a morbidity rate of 115.1 per 100,000 of population and North Dakota one of 88.7. The incidence was also high in Wyoming and in Colorado where the morbidity rate was 80.9. The Middle and South Atlantic states had the lowest incidence as a whole. There has been a mounting increase in the ratio of cases reported to deaths in recent years. Thus in the period 1930-34 there were only 6.8 cases reported per death, while in 1946 the ratio was 13.3 per death. It seems probable that the increase in the number of cases is due in part at least to an increasing reporting of nonparalytic cases. Up to June 21 of this year (last figures available) the number of cases reported since the low seasonal week for the disease (March 15-21) was 513 as contrasted with 1,112 in 1946. California was the only state showing an increasing number of cases.

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At the annual meeting of the **Section on Pediatrics of the American Medical Association** at Atlantic City in June, the following officers were elected for 1948: Chairman, Dr. Oscar Reiss, Los Angeles; Vice-Chairman, Dr. Stanley Gibson, Chicago; and Secretary, Dr. M. M. Nicholson, Washington, D. C.

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THE HOSPITAL ACT AND YOUR COMMUNITY

If you are interested in the hospital program, authorized last year by the Hospital Survey and Construction Act, this booklet will tell you, in simple terms, what the Act means to you, your community, and your State. It should be especially valuable to any group or community that wants to build a hospital but does not know how it goes about getting Federal aid for construction. Single copies are available free on request to the U. S. Public Health Service, Washington 25, D. C.

# Book Reviews

**Child and Adolescent Life in Health and Disease. A Study in Social Paediatrics.** W. S. Craig, Baltimore, 1946, Williams and Wilkins Co., 667 pages. Price $7.00.

It is sincerely to be hoped that someone will do for the United States what Dr. Craig of Edinburg has done for Great Britain in this volume of "social pediatrics." In Part I he has traced the historic background and development of the many measures which have been developed over the years for the care and protection of the child. Some of the effort has been due to voluntary agencies and philanthropy, and some to governmental action. The most striking thing is the myriad of agencies which have developed and now exist to take care of some one need or interest, with a resulting overlapping of function and a most confusing present-day picture when looked upon as a whole. The same situation exists in our own country, but it is doubtful if the means and mechanisms which are available today to protect and insure the health of the child could have been developed in any other way. The book shows unending research. In Part I, the work and part played by voluntary agencies, the care of the dependent and defective child, the development of hospitals and medical care, school hygiene, and infant and child welfare are traced from their earliest beginning to their present-day status.

In Part II, the author presents the care of child life at the present time, the laws for protection of the child, and the present-day agencies for maintaining health, and the care of the sick and handicapped child. To this he has added a most interesting chapter on the child under conditions of total war which discusses the problems and measures taken to protect the children of Great Britain in World War II. This summary of the problem of evacuation is most important and should be read by all who might, at some future time be faced with a similar problem. Part III is a brief discussion of the future, in which the author stresses the need for research on a broad basis and the importance of child hygiene in medical education. Part IV is in the nature of an appendix discussing the various laws relating to child health and welfare in Great Britain.

It is a book not only of interest and importance to the pediatrician, but equally to the social worker and public official. It is a most unique book in that while specific phases of child care have been reviewed and presented in a monograph form, for the first time so far as we know, the attempt has been made to present the picture of child care as a whole.

B. S. V.

**The Personality of the Preschool Child.** Werner Wolf, New York, 1946, Grune & Stratton, 341 pages. Price $5.00.

The personality of the preschool child has interested educators and psychologists more than physicians. Even contributions from psychiatrists about this age period are meagre. This book is the work of a psychologist, who, as an educator and researcher, has a special interest in the emotional development of children.

The book is made up of three parts. Part One deals with material which was obtained by direct observation of children in the setting of nursery school and home. In this section one finds much interesting and valuable information about the mind of the child and how it functions, what the language of his emotions are and in what the child's concept of reality consists. There is mention of such important items as dreams, play, and social life. The reader will find discussions of the topics of birth and death which every healthy child sooner or later presents to his parents and teachers. In Part Two, the author deals with his

experiments in studying the preschool child as an individual with feelings and with intelligence. Various new techniques of studying the projection of feelings through play and art are described. Part Three deals with theory about principles of children's art and the development of a child's personality. It contains a chapter on research methods in child study which should interest particularly the scientifically trained pediatrician who has so long decried the unscientific research techniques of the psychologist and psychiatrist. Following this is a bibliography of 628 publications. This book will furnish much useful information to the practitioner of pediatrics who reads it in small bits when stimulated to seek information about topics which are presented in the everyday care of preschool children. For the pediatrician with psychiatric experience and for the individual doing research in child development, the book has still greater value not only as a source book, but also as a bibliographical reference.

SENN.

# Editor's Column

## THE CANADIAN MEDICAL ASSOCIATION AND HEALTH INSURANCE

The relationship between medicine in Canada and medicine in the United States has always been close. The medical schools of both countries have the same Scotch-English heritage. The general scheme of medical practice and hospital organization is quite the same. Many outstanding leaders, as Osler, have taught in both countries, and hundreds of our younger physicians have interchanged for their education and training. Nearly all of our special societies include Canadians in their membership, and frequently these societies meet in Canadian cities. The two large national societies, the Canadian Medical Association and the American Medical Association, are quite similar in their character and plan of organization. As a result there exists a close interchange of medical thought between the two countries as well as close friendships between members of the profession.

In light of this close relationship and common interests the "Submission Respecting Health Insurance by the Canadian Medical Association" to the "Special Committee on Social Security" of the Canadian House of Commons, which is reprinted in Dr. Park's column on "The Social Aspects of Medicine" in this issue (p. 228) gives food for much thought. Exactly the same problems are facing medical practice in Canada as exist here in the United States: problems of the distribution of good medical care, problems of making good medical care available to all, problems of the increasing cost of good medical care due to technical and scientific advances, problems of hospitals, problems of medical education. In view of the similarity of the problems facing medicine in the two countries, the attitude of the medical profession in Canada, as represented by their national association, is not only of interest but importance. Throughout the ten years prior to the "Submission" in 1943, the Canadian Medical Association consistently sought to ascertain the opinion of the medical profession on the subject of health insurance, and the "Submission" crystallizes these opinions which are presented in the report.

It should be read in its entirety by all our readers, who, whether they are interested or not, are involved in this pressing question of health insurance. In the Canadian report, the proposals of the Canadian Association are clear, simply stated, liberal, and full of common sense.

With this attitude of help and cooperation on the part of the Canadian Medical Association in assisting the Canadian government in solving the complicated problem of medical care for its citizenry, we are satisfied that the interests of the Canadian practitioner will be fully protected in whatever legislation is finally promulgated.

## THE FIFTH INTERNATIONAL CONGRESS OF PEDIATRICS

There were many, including the writer, who had serious doubts as to the advisability of holding the Fifth International Congress of Pediatrics this year. Due to the war, it has been postponed for several years, and to many of us the wise course seemed to put it off until world conditions become more settled. We were wrong, and those who urged the Congress at this time were right. It was an outstanding success. An old friend from one of the war-torn countries tried to tell me what it meant to him to attend a meeting with his pediatric colleagues and friends from all over the world, after years of scientific and medical isolation, and to see developments of which he knew little. "I feel as if I had been born again and am going back filled with the desire to work once more."

All the time the meeting was going on with the closest friendship and feeling among the representatives from over sixty countries, the newspapers were filled with stories of distrust and threats of war throughout the world. A meeting such as the Fifth Congress makes one wonder if it is not possible in some way to spread the religion of medicine and the feeling of physicians toward their work and toward one another into other spheres. If this could be done, there would be no distrust or talk of war.

Certainly in times such as these, of world turmoil and trouble, the holding of the Congress shows there is a background of peace and decency left in the world.

B. S. V.